Atypical Celiac Disease Resistant to Thyroxine Replacement

Tiroksin Replasman Tedavisine Dirençli Atipik Çölyak Hastalığı

Çölyak Hastalığı, Hipotiroidi, Levotiroksin / Celiac Disease, Hypothyroidism, Levothyroxine

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Özet

Çölyak hastalığı gluten içeren diyet alımına bağlı olarak gelişen immün aracılı enteropatidir, özellikle otoimmün tiroid hastalığı gibi diğer endokrin hastalıklarla yakından ilişkilidir. Çölyak hastalığı ve özellikle hashimato tiroiditine sekonder hipotiroidi birlikteliği literatürlerde sıklıkla vurgulanmaktadır. Çölyak hastalığı ve otoimmun tiroid hastalığı arasındaki ilişki ilk olarak üç dekad önce tanımlanmıştır. Hastalar genellikle steatore ve diyare gibi klasik bulgularla başvurabilir fakat artan L-tiroksin dozu ihtiyacı ve kilo kaybı hipotiroidizim ile birlikte çölyak hastalığını iyi tanımlamaktadır. Bu olguların tiroksin replasmanına dirençli olduğu bilinmektedir. Biz 35 yaşında atipik çölyak hastalığı olan ve 1600 mcg/gün gibi oldukça yüksek doz tiroksin replasmanına ihtiyaç duyan kadın olguyu sunuyoruz.

Anahtar Kelimeler

Çölyak Hastalığı; Hipotiroidi; L-Tiroxin Replasmanı

Abstract

Celiac disease, an immune-mediated enteropathy that develops in susceptible individuals upon ingestion of gluten containing diet, is closely associated with other autoimmune endocrine disorders, particularly autoimmune thyroid disease. Celiac disease and hypothyroidism (especially due to Hashimoto disease) cooccurence is frequently mentioned in the literatüre. The relationship between celiac disease and autoimmune thyroid disease was first described three decades ago. Patients usually have the classical presentation of diarrhoea and steatorrhoea but hypothyroidism with weight loss and increased dose requirement of L Thyroxine are two well recognised presentations of celiac disease in hypothyroidism. It is known that these cases are resistant to thyroxine replacement. Herein we presented a 35 year old female patient with atypical celiac disease and needed an extremely high dose of thyroxine such as 1600 mcg/day for treatment.

Keywords

Celiac Disease; Hypothyroidism; L-Tiroxin Replacement

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Introduction

Hypothyroidism is a common endocrine disorder that mainly affects women and the elderly [1]. Celiac disease (CD) is an immune-mediated disease triggered by an environmental agent, gluten, in genetically predisposed individuals, characterized by villous atrophy of the proximal small intestine and malabsorption. The association between CD and other autoimmune diseases such as type 1 diabetes mellitus, autoimmune thyroiditis, and other endocrine diseases has been reported in many studies in both children and adults [2]. Celiac disease and hypothyroidism (especially due to Hashimoto disease) cooccurence is frequently mentioned in the literature [3]. It is known that these cases are resistant to thyroxine replacement [4].

Case Report

35 year old female patient was referred due to complaints of weight gain, facial and periorbital swelling, constipation, and irregular menstrual cycle. In her medical history, she was operated for euthyroid multinodular goiter (EMG) in another center and the postoperative pathological analysis were benign. Levothyroxine (LT4) replacement was initiated during the postoperative period, but weekly dose was increased 50 mcg due to persistent high TSH levels. When the patient was referred to our clinics, she was taking a LT4 therapy of 1600 mcg/day and had complaints of forgetfullness, irregular mensturation, weight gain, and constipation. In addition LT3 therapy has not been initiated so far. Her physical examination showed an blood pressure of 110/70 mm/Hg, periorbital edema and decrease in intestinal movements. Patient was regularly taking her meds before meals and was not on any other drug or herbal supplement. Her laboratory values were as follows; fasting blood glucose: 90 mg/dl (74-106), creatinine: 0.83 mg/dl (0.6-1.3), albumin: 4.5 gr/dl (3.5-5.2), Hgb:13.7 gr/dl (13.6- 17.2), Ca: 9.1 mg/dl (8.8-10.6), P: 2.8 mg/dl (2.5-4.5), Fe: 113 mg/dl (60-180), Ferritin: 5.3 ng/ml (13-150), 25(OH)D3: 7.1 ng/ml (20-100), Vit B12:382 pg/ml (191-663), TSH: 15 µIU/ml (0.34-5.6) and cortisol: 34 mcg/dL. Malabsorbsion was thought as a cause and relevant investigations were carried out. Abdominal ultrasonography was within normal limits. Celiac antibodies (Anti-endomysium, anti-gliadin and tissue transglutaminase) were negative. Upper gastrointestinal endoscopy was performed; antral erosive gastritis and a flaccid lower esophagus was observed. Biopsies were taken from antrum, corpus and the second segment of duodenum. Biopsies of antrum and corpus revealed activated chronic gastritis, helicobacter pylori presence, and intestinal metaplasia. Intraepithelial lymphocyte increase (modified Marsh classification Type 1) was detected in duodenum biopsy (figures:1-2). Following gastroenterology consultation, patient was diagnosed as having atypical gluten enteropathy and a gluten free diet was initiated. Patient complied well with the diet and in her follw ups the LT4 dose was decreased to 600 mcg which is the dose the patient is currently on. Lowest TSH recorded in a year of follow up is 8 µIU/ml.

Discussion

Cooccurence of celiac disease and autoimmune hypothyroidism is frequent [4]. Our patient was unique in the sense that she did not possess autoimmunity and was a postoperative hypothyroidism case operated due to EMG. Recently, increases in the prevalance of atypical celiac disease of the adult are reported [5]. Atypical presentation is characterized by scarce clinical symptoms. Frequently, only a single symptom is observed, such as lack of body mass increase and growth retardation, anemia, dental enamel hypoplasia, osteoporosis, or pubertal delay. Silent presentation of celiac disease is diagnosed in asymptomatic patients with increased risk of the disease. Among diseases and conditions [6]. Our patient had no signs of malabsorbtion and had negative celiac antibodies, thus the diagnosis was histopathological. Various absorbtion disorders secondary to gastroenterological pathologies are common [7]. However, we find it difficult to explain such an extraordinary thyroxine resistance in our non autoimmunized patient only by the presence of atypical celiac disease. We believe problems with the pharmacokinetics of the drugs may be responsible for this outcome. Some reports in the literature point to suboptimal thyroxine replacement in a respectable proportion of the population [8]. As a result; after malabsorbtive causes are excluded, adequacy of the replacement and possible thyroxine resistance should be addressed.

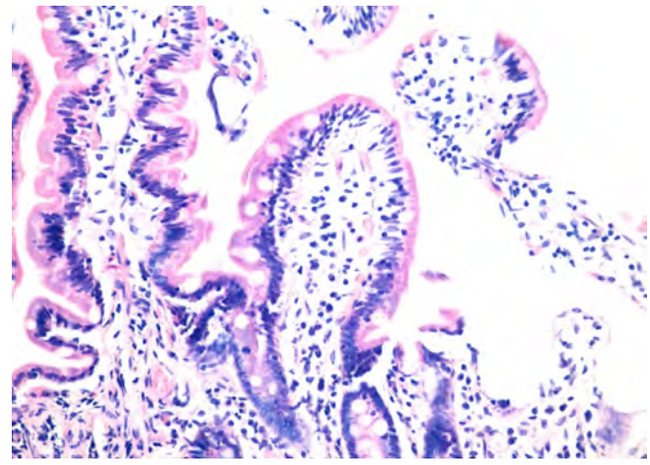


Figure 1. Intraepithelial lymphocyte infiltration in duodenum biopsy (H&E, x100).

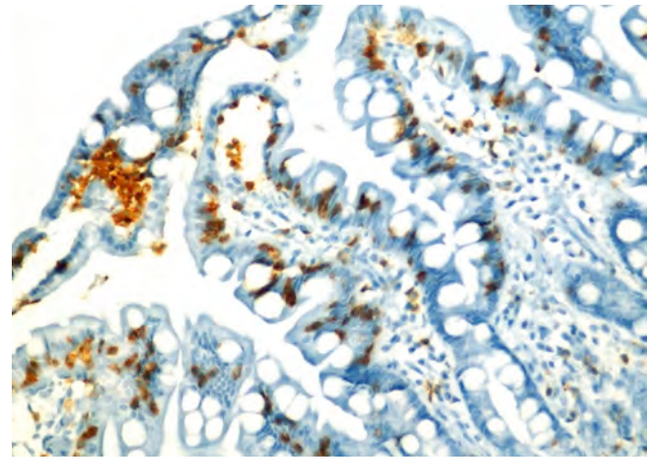


Figure 2. Immunohistochemical CD3 positivity in the intraepithelial lymphocytes (CD3, x200).

Competing interests

The authors declare that they have no competing interests.

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