ARTICLE

Co-Occurrence of Celiac Disease and Ulcerative Colitis in a 12-Year-Old Girl

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Celiac disease (CD) and inflammatory bowel diseases (IBD) are separately well-described entities, but the co-occurrence in children has been very rarely reported until today. According to the literature, this case about 12-year-old girl would be the fifth case ever published about co-occurrence in children. We presume that there should be a higher comorbidity prevalence than that described. Distinguishing both diseases in one patient could be difficult due to the overlapping symptoms, but it is very important considering completely different therapeutic approaches.

Keywords: celiac disease, IBD, ulcerative colitis, co-occurrence, genetics

INTRODUCTION

Celiac disease (CD) and inflammatory bowel diseases (IBD) are separately well described entities. CD is one of the most common lifelong disorders, affecting approximately 1% of the general population [1, 2]. The comorbidity between CD and other autoimmune disorders has been clearly established [3]. However, the associated occurrence of CD and ulcerative colitis (UC) in children has been very rarely reported until today, referring only to four case reports with confirmed diagnosis [4]. Distinguishing both diseases in one patient could be difficult due to the overlapping symptoms, but it is very important considering completely different therapeutic approaches.

We report our experience in a 12-year-old girl with co-occurrence of CD and UC.

CASE REPORT

A 12-year-old girl was admitted due to chronic diarrhea, looking chronically ill and seriously underweight (BMI 12.6 kg/m², Z-score -3.7). She complained about diarrhea lasting for 3 weeks, nausea without vomiting and loss of appetite. Laboratory testing revealed anemia, mildly elevated sedimentation rate (27 mm/h) and C-reactive protein (6.3 mg/l), lower albumins (32.8 g/l) and very low serum vitamin D3 levels

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(23 nmol/l). The benzidine stool test was positive. The results of other laboratory tests were in reference intervals. Infective etiology was excluded.

Considering the high values of IgA anti-tissue transglutaminase antibodies (anti-TG2; 163.3 RU/ml), and IgG antibodies to deamidated gliadin (anti-DGP; 41 RU/ml), we performed an esophagastroduodenoscopy (EGD) along with the duodenal biopsy that revealed histopathological changes typical for CD (Marsh IIIA) (Figure 1). A gluten-free diet (GFD) was initiated. In the period that followed bloody, loose stools appeared. Repeated microbiological testing re-excluded infective etiology. Colonoscopy was performed due to the significantly elevated fecal calprotectin (FC, 1296 μg/g). The endoscopic, as well as the histopathological findings of colon were typical for UC (Figure 2). MR enterography findings confirmed the UC. After the definitive diagnosis of UC (Pediatric UC Activity Index – PUCAI score 60), along with the CD, prednisone and mesalazine therapy was started, and GFD was continued.

The patient reacted rapidly to the treatment. Her stools normalized in a few days. She gained some weight and strength. At discharge her clinical feature was normal (PUCAI score 5). The clinical course in the following 12 months showed excellent physical condition of the patient. After corticosteroid discontinuation, there were no signs of UC relapse, so we continued only with 5-ASA and a GFD. Follow-up laboratory testing revealed a significant decrease of anti-TG2 levels (50.7 RU/ml), and normalization of all laboratory findings. The patient continued to gain weight and on her last visit her body weight was 43.2 kg (BMI 18.1 kg/m²; Z-score −0.25).

DISCUSSION

In 2005, when Yang [5] published the first database cohort study that referred to co-occurrence of CD, and IBD in adult patients, there have been only a few isolated
Figure 2. Colon mucosa; distortion of crypt architecture, inflammation of crypts (cryptitis), frank crypt abscesses and inflammatory cells in the lamina propria (H&E 100×), inset: depletion of surface mucous secretion (H&E 200×).

reports about this comorbidity. Results of the study showed that in 10 out of 455 patients with CD, IBD was confirmed (5 UC and 5 Crohn’s disease). They proved that IBD is more common in patients with CD than in the general population.

The first case report regarding the associated occurrence of CD and UC in children was published in 1999 by Day and Abbott [6]. They described a 7-year-old New Zealand girl with simultaneous presentation of CD and UC with severe clinical course. Over an 18 month following discharge, she required frequent courses of corticosteroid therapy and initiation of azathioprine in addition to ongoing mesalazine therapy. In 2004, Sykora [7] described a case of a 15-year-old patient with simultaneous presentation of CD, UC and autoimmune thyroiditis and favorable clinical course over a 1-year follow-up period. The third case was different. A 7-year-old boy was diagnosed with CD, UC and primary sclerosing cholangitis virtually simultaneously, which led to a more aggressive phenotype of IBD, that in turn required an immunomodulatory therapy [8]. The fourth case report, published by Cheng [9] described an 8-year-old boy with a history of anemia and failure to thrive who presented with bloody diarrhea. Eventually, CD and UC colitis were diagnosed. This male patient as well as, the female patient in our case report, had a benign disease course on a GFD and mesalamine therapy for 2 years after corticosteroid tapering with no need for immunomodulators.

In our case, the patient who was diagnosed with CD had high levels of FC, accompanied with persisting diarrhea, regardless of following a GFD. This led us to suspect an IBD associated disease. Following a confirmative colonoscopy diagnosis of UC, we prescribed prednisone and mesalazine therapy in continuation of GFD. A rapid response to the therapy was observed through normalization of laboratory tests and stools, weight gain and a favorable clinical course even after the discontinuation of
corticosteroid therapy. There was no need for immunomodulatory therapy over the 12 months following discharge.

Recently, a case-control study conducted at two tertiary referral hospitals in the USA among adults with CD and IBD has been published. Results showed that pancolitis was more common in celiac – UC patients when compared with controls, with a trend towards increased use of immunomodulators. On the other hand, coexisting CD did not influence natural history of Crohn’s disease. These results suggest more aggressive phenotype in case of CD and UC co-occurrence [4].

In conclusion, due to the high incidence of each of these diseases, as well as possibility of shared genetic risks, we assume there should be higher comorbidity prevalence than those described in the literature [10]. Further studies are required to assess the prevalence of this association in children. Whether the shared genetic risk between both diseases predisposes a more aggressive phenotype of IBD remains to be seen.

We hope this report will bring new cases to light and give us more information about phenotype differences in cases of CD and UC co-occurrence which would lead to further discussions about optimal diagnostic and therapeutic approaches.

Declaration of Interest

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

REFERENCES


