Celiac disease, gluten-free diet and health-related quality of life

Celiac disease (CeD) is an immune-based systemic condition triggered by gluten in genetically predisposed individuals, and characterized by the presence of a variable mixture of gluten-dependant clinical manifestations, specific antibodies, haplotype HLA-DQ2 or HLA-DQ8, and bowel disease (1). Population-based screening studies show it to be an underdiagnosed disease with a seroprevalence of about 1%-2% amongst our population. The clinical expression of CeD is highly variable, and oscillates from severe forms with diarrhea, steatorrhea, and wasting (classical presentation) to oligosymptomatic or asymptomatic illness. In the adult atypical presentations with nonspecific gastrointestinal and/or extraintestinal manifestations are increasingly common (2). Some gastrointestinal symptoms, including dyspepsia, recurrent abdominal pain and diarrhea, are highly prevalent in the adult, and may be erroneously attributed to some functional digestive disorder (3). CeD may also induce psychological changes mainly involving the positivity, wellbeing, anxiety and vitality axes, with severity paralleling gastrointestinal symptoms (4). The therapy recommended to achieve symptom control and avert complications is a lifelong, stringent gluten-free diet (GFD) (2). Interest in the assessment of health-related quality of life (HRQoL) in CeD has increased of late, allowing a better understanding of the condition and an additional tool for patient follow-up, besides clinical response and laboratory parameters. On the other hand, patient attitudes and expectations may have a substantial impact on GFD compliance, hence insight into the subjective perceptions and desires of patients is particularly important in a disease requiring permanent lifestyle modifications (5).

When an individual is diagnosed with CeD he or she must face relevant long-term changes. On the one hand, the development of disease-derived symptoms entails a biological impact. On the other, the perception of being ridden with a chronic illness, the need to adhere to a restrictive, demanding, permanent diet, regular medical follow-up, and the possibility of relatives sharing the disease determine a relevant psychosocial impact. In adults, a diagnosis with CeD entails lifestyle changes due to the development of emotional disturbances including feeling different from others, shame or fear of contamination with gluten, and the additional difficulties of GFD adherence when not at home, particularly at work or on a trip (6). Several studies have shown that all these bio-psycho-social changes have a negative impact on the HRQoL of patients with CeD. Two studies in collaboration with national celiac associations, one in Germany, the other in Canada, where a generic questionnaire was mailed to 1,000 and 5,240 patients, respectively, confirm the hypothesis that celiac patients, when compared to the general population, have poorer scores in all the assessed quality of life dimensions (7,8). In our setting, Casellas et al. (9) published a paper in 2005 where, using the generic EuroQol questionnaire and the Spanish version of the Gastrointestinal Quality of Life Questionnaire (GIQLI), it was shown that patients with newly diagnosed CeD had a poorer HRQoL. This study highlights the impact of CeD on all the assessed dimensions, including those related to symptoms, and the emotional, physical and social domains. Several factors may play a role in the declining HRQoL of celiac subjects (10,11). One is the presence of symptoms or comorbid conditions, as patients with more manifestations complain of poorer HRQoL. Another relevant factor is diet adherence, such that celiac individuals with dietary non-compliance or erratic adherence score significantly worse in all the dimensions included in the generic SF-36 questionnaire. As with other chronic intestinal conditions, the impact of disease on HRQoL is more pronounced in females. Casellas et al. (12), in a another study also assessing HRQoL among 340 patients with CeD (163 of them on GFD) using the generic EuroQol questionnaire and the Spanish version of GIQLI, confirmed these results in our setting. These authors, in a subsequent study (13), showed how fatigue, as assessed with a specific questionnaire, is an additional factor associated with poorer HRQoL among patients with untreated CeD.

Fortunately enough, this impairment of HRQoL improves, as do symptoms, with diet therapy. Green et al. (14) mailed some generic questions on perceived HRQoL, both before and after diagnosis, to an extensive series of patients from associations and internet forums. Of all 1,612 patients who responded, 63% claimed a modest or poor HRQoL before diagnosis. Among these, HRQoL significantly improved for 77% following diagnosis and treatment. As previously described, full adherence to GFD is also a determinant of improved HRQoL. (12). Despite all this, a non-negligible proportion of celiac subjects exhibit inadequate compliance with their diet. A study in our setting, again by Casellas et al. (15), has shown that therapy is usually complied with by most patients, with intentional non-compliance amounting to 1.8% and unintentional non-compliance (from carelessness or forgetfulness) to 15.5% of cases. Several factors may be involved in poorer diet compliance, including the unattractive looks of gluten-free products, eating outdoors, no symptoms, difficulties with food labels, or a hard time finding gluten-free products. Because of these reasons, it is no wonder that some available studies show conflicting results as regards the benefits of GFD on HRQoL. Thus, patients with silent or asymptomatic CeD who are diagnosed with serology may experience HRQoL impairment.
on GFD (16). Several studies using generic questionnaires such as the SF-36 have shown that the extent of GFD compliance not always is associated with improved HRQoL, and the difficulties encountered by patients to stringently adhere to GFD may also have a negative impact on HRQoL (17,18).

In the paper published in the present issue of the Revista Española de Enfermedades Digestivas (Spanish Journal of Gastroenterology), authors Casellas et al. (19) provide new evidence for the benefits of adequate compliance with GFD on the HRQoL of adult patients with CeD. In this interesting prospective, multicenter study involving 7 hospitals both generic and specific HRQoL questionnaires were administered to a total of 366 adult patients with CeD on GFD for at least 1 year. Adherence to GFD was assessed using Morisky’s questionnaire, in this case adapted to assess diet compliance with an additional answer: “I never forget my diet”. A total of 71.5% of patients exhibited a strict compliance with GFD, 23.5% had unintentional non-compliance, and 5% had intentional non-compliance. These values are similar to those previously published, which describe adequate compliance in 70%-81% of adults with CeD. The results of HRQoL assessment show how perfect adherence to GFD, good symptom control, and considering stringent GFD an easy task was significantly associated with better scores in quality of life questionnaires. A relevant aspect to be highlighted is that, in contrast with most prior studies, the authors here use a specific questionnaire—namely the CD-QOL questionnaire, in addition to the generic EuroQOL-5D—to assess HRQoL. The CD-QOL questionnaire was specifically designed for the assessment of HRQoL in patients with CeD; it has also been recently translated and validated in our setting by the authors of the present paper (20). Casellas et al. also provide new evidence on the extent of HRQoL recovery after treatment with GFD. While GFD, as previously described, improves HRQoL in people with CeD, there is no unanimity on whether recovery is complete—reaching the values of the healthy population—or partial. As the CD-QOL dimension with poorest patient scores is the one related to “inappropriate therapy”, the authors conclude that the absence of therapies alternative to GFD reduces the HRQoL of patients as compared to a healthy population. A study limitation, as the authors themselves point out, is the absence of HRQoL measurements before GFD onset. This would have allowed to determine the extent of the perceived health gain as related to diet adherence using a specific questionnaire.

The present study of HRQoL in CeD is a significant contribution to our understanding of the aforementioned disease and of how patient attitude and expectations may play a role in adequate compliance with GFD despite its arguable benefits for symptom control and HRQoL. One of the study’s strengths is its underscoring the significance of validated instruments to objectively measure the response to gluten suppression in all dimensions (physical, emotional, social), particularly when symptom remission is a diagnostic criterion as posited by Catassi et al. (21), for whom symptom response and enteropathy regression represent a requirement for seronegative forms (4 out of 5 rule).

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REFERENCES


