



# Hypervigilance to a Gluten-Free Diet and Decreased Quality of Life in Teenagers and Adults with Celiac Disease

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## Abstract

**Background and Aims** Avoidance of gluten is critical for individuals with celiac disease (CD), but there is also concern that “extreme vigilance” to a strict gluten-free diet may increase symptoms such as anxiety and fatigue, and therefore, lower quality of life (QOL). We examined the associations of QOL with energy levels and adherence to, and knowledge about, a gluten-free diet.

**Methods** This is a cross-sectional prospective study of 80 teenagers and adults, all with biopsy-confirmed CD, living in a major metropolitan area. QOL was assessed with CD-specific measures. Dietary vigilance was based on 24-h recalls and an interview. Knowledge was based on a food label quiz. Open-ended questions described facilitators and barriers to maintaining a gluten-free diet.

**Results** The extremely vigilant adults in our sample had significantly lower QOL scores than their less vigilant counterparts [(mean (SD): 64.2 (16.0) vs 77.2 (12.2),  $p = 0.004$ ]. Extreme vigilance was also associated with greater knowledge [5.7 (0.7) vs 5.1 (0.8),  $p = 0.035$ ]. Adults with lower energy levels had significantly lower overall QOL scores than adults with higher energy levels [68.0 (13.6) vs 78.9 (13.0),  $p = 0.006$ ]. Patterns were similar for teenagers. Cooking at home and using internet sites and apps were prevalent strategies used by the hypervigilant to maintain a strict gluten-free diet. Eating out was particularly problematic.

**Conclusion** There are potential negative consequences of hypervigilance to a strict gluten-free diet. Clinicians must consider the importance of concurrently promoting *both* dietary adherence and social and emotional well-being for individuals with CD.

**Keywords** Celiac disease · Quality of life · Adherence · Gluten-free diet · Teenagers · Adults

## Abbreviations

CD Celiac disease

CDAT Celiac disease adherence test

DF Degrees of freedom

NJ New Jersey

NY New York

NYC New York City

QOL Quality of life

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RDN	Registered dietitian nutritionist
SDE	Standardized dietitian evaluation
GF	Gluten free

## Introduction

Celiac disease (CD) is an autoimmune disorder featuring duodenal villous atrophy triggered by dietary gluten, a protein present in wheat, barley, and rye. CD affects multiple systems in the body and can manifest with a variety of symptoms, including diarrhea, abdominal pain, peripheral neuropathy, anemia, and infertility [1, 2]. The prevalence of CD has increased up to fivefold in the United States since 1950, and diagnosis rates continue to rise, a consequence of parallel trends of increased prevalence and improved awareness and testing [3–5].

Current guidelines for the management of CD specify lifelong adherence to a strict gluten-free diet [6, 7]. For the majority of patients, strict avoidance of gluten is the only treatment proven to result in clinical, serologic, and histologic improvement [8–10]. Observational studies using duodenal mucosal healing as a marker suggest that adherence to a gluten-free diet can decrease risk of long-term complications such as osteoporotic fracture and lymphoproliferative malignancy [11, 12].

Since the level at which gluten is harmless is not known for the individual with CD, avoidance of all gluten (from food, beverages medications, and supplements) is the current standard of care [6, 7]. Managing such a restrictive diet is challenging, and treatment burden can be high [13–17]. Individuals with CD must learn what foods to eat, what foods to avoid, hidden sources of gluten, and how to navigate a complex food environment and a lifetime of social situations.

While better adherence to a gluten-free diet has been associated with better quality of life (QOL) [18–22], there is also concern that “extreme vigilance” to a gluten-free diet may increase symptoms, such as anxiety and fatigue, and, therefore, lower QOL. In other words, there may be a cost to hypervigilance for some individuals with CD when following a strict gluten-free diet.

This study examines the associations of CD-specific QOL with energy level and adherence to, and knowledge about, a gluten-free diet. We also explore specifics of how QOL may be affected by describing qualitative barriers and facilitators to maintaining a gluten-free diet. Our purpose is to inform future nutrition education strategies which can promote a strict gluten-free diet while helping to maximize QOL for teenagers and adults with CD.

## Methods

### Design

We performed a cross-sectional prospective study of adult and teenaged patients with CD, focusing on QOL and its correlates. The study was approved by the Institutional Review Boards at the Columbia University Medical Center and at Teachers College Columbia University.

### Setting and Participants

The study was conducted at the Celiac Disease Center of Columbia University in New York City. Inclusion criteria required that participants be at least 13 years of age, self-report a duodenal biopsy-confirmed diagnosis of CD  $\geq 1$  year prior, and be willing to participate in three visits (one in person and two via telephone) over a 1-month period. We considered 13–17 year olds to be teenagers and those 18 years and older to be adults. Exclusion criteria included serum or self-diagnosed CD (without biopsy), a CD diagnosis  $< 1$  year prior, and age  $< 13$  years old. The teenagers received a \$25 Amazon gift card for their participation.

### Enrollment

Enrollment occurred between March and August 2016. Our target goal was 30 adults and 30 teenagers. Enrollment exceeded our expectations. All affiliates (~ 5000 members) of the Celiac Disease Center of Columbia University (which includes a mix of patients, family members, and those with an interest in CD) were emailed initially asking about their interest in the study. Two additional follow-up emails specifically targeted teenagers. Those interested were assessed for eligibility by telephone. Among the 123 respondents to the email invitation (78 adults; 45 teenagers), 43 were ineligible (28 adults, 15 teenagers) and 80 were eligible and enrolled (50 adults and 30 teenagers). Among the 28 ineligible adults, 6 were ineligible due to not having a duodenal biopsy to confirm CD, 20 for never scheduling an appointment, and 2 for other reasons. Among the 15 ineligible teenagers, 11 were ineligible for not having a duodenal biopsy to confirm CD, 3 for never scheduling an appointment, and 1 for other reasons.

### Data Collection and Measures

#### Demographic and Medical History Variables

Age (date of birth), gender (male, female), self-described ethnicity (Hispanic, non-Hispanic), self-described race (White, African-American, Asian, Other), education

(highest level/grade achieved), and home residence (based on zip code) were assessed. Medical history variables included years since CD diagnosis, affiliation of their gastroenterologist (Celiac Center of Columbia University vs Other), and visits with a registered dietitian, now referred to as a registered dietitian nutritionist (RDN) (currently, once only, more than once, never).

## Celiac Disease-Specific Quality of Life

### CDQOL

CD-specific quality of life (CDQOL) in adults was assessed using a 20-item validated survey instrument [23]. Participants answered questions with Likert scales where 1 = not at all, 2 = slightly, 3 = moderately, 4 = quite a bit, and 5 = a great deal. Answers were transformed and combined to obtain an overall score and four clinically relevant subscales: dysphoria (4 items), limitations (9 items), health concerns (5 items), and inadequate treatment (2 items). Dysphoria items measured the extent to which individuals feel depressed, frightened, or overwhelmed by CD. Limitation items measured the extent to which individuals feel limited by CD when eating out with others, socializing, and traveling. Health concern items measured the extent to which individuals feel worried about long-term health outcomes of CD for themselves or other family members. Inadequate treatment items measured the extent to which individuals feel there are enough treatment options for their CD. Each final score had a possible range of 0–100 with higher scores suggesting a higher degree of QOL.

### CDPQOL

CD-specific pediatric quality of life (CDPQOL) in teenagers was assessed using a 17-item validated survey instrument [24]. Participants answered questions with Likert scales where 0 = never, 1 = almost never, 2 = sometimes, 3 = often, 4 = almost always. Answers were transformed and combined to obtain an overall score and four clinically relevant subscales: social (7 items), uncertainty (3 items), isolation (4 items), and limitations (3 items). Social items measured self-esteem and the extent to which individuals feel they are not understood or a burden. Uncertainty items measured the extent to which individuals are worried about college, their future, and getting older with CD. Isolation items measured the extent to which individuals feel different from their family and friends because of their CD. Limitation items measured the extent to which individuals avoid parties or feel nervous about eating at a friend's house. Each final score had a possible range of 0–100 with higher scaled scores suggesting a higher degree of QOL.

## Dietary Adherence and Vigilance

Dietary adherence was assessed using the Standardized Dietitian Evaluation (SDE) instrument [25] which utilizes evaluations from trained Masters students in nutrition. Three 24-h dietary recalls collected over a 1-month period and an interview were reviewed for quantity and frequency of gluten exposure (e.g., uses celiac-friendly restaurants or asks thorough questions when dining out, has eliminated cross-contamination potential in kitchen.) each recorded on a 6 point Likert scale ranging from 1 (excellent adherence) to 6 (not currently following a gluten-free diet).

Participants were divided into two groups: the “extremely vigilant” and the “less vigilant.” Participants that received an excellent adherence score for all 3 days of 24 h dietary recalls were considered to be “extremely vigilant” (i.e., only scores of 1 for all categories). All others were considered to be “less vigilant” (scores of 2–6 for any of the categories on any of the 3 days of 24 h dietary recalls).

### Energy Level

One item from the Celiac Disease Adherence Test (CDAT) [25] was used to classify participant's energy level. Participants were asked the extent to which they were bothered by low energy over the past 4 weeks with 1 = none of the time, 2 = a little of the time, 3 = some of the time, 4 = most of the time, and 5 = all of the time. Those who responded  $\geq 3$  were considered to have “lower energy” and those who responded 1 or 2 were considered to have “higher energy.”

### Knowledge

Knowledge about gluten-containing ingredients was assessed with a food label quiz developed by Leffler et al. [25]. Participants were shown a modified food label and asked to identify ingredients that contained or possibly contained gluten. Individuals received 1 point for each of the 6 (out of 22) ingredients correctly identified as potentially containing gluten. Scores could range from 0 (lower knowledge) to 6 (higher knowledge).

### Facilitators and Barriers

To assess barriers, participants were asked, “What do you see as the major challenges to following a strict gluten free diet?” To assess facilitators, participants were asked, “What do you see as the things that help make it easy to follow a strict gluten free diet?” Interviewers allowed participants to spontaneously report whatever came to mind and then probed to elicit further detail. All handwritten notes were analyzed for major themes according to the methodology described by Braun and Clarke [26]. Sample responses were

selected as representative of each theme. Some common themes were apparent immediately (e.g., dislikes having to ask questions, worrying about cross-contamination). Others emerged after multiple readings. Some themes, not necessarily common, were highlighted because of extreme specificity (e.g., lack of labels for medications, makeup as a barrier; other family members with CD as a facilitator).

## Statistical Analysis

Means and standard deviations are presented for continuous data, frequencies, and percentages for categorical data. T-tests were used to assess group differences in QOL, knowledge, and/or adherence between those with higher versus lower energy levels and those that were extremely versus less vigilant in following a strict gluten-free diet. Barriers and facilitators are described with sample responses. Chi-square tests with continuity correction were used to assess group differences in frequency of barrier and facilitator themes.

Adults and teenagers were analyzed separately. We considered  $p < 0.05$  as statistically significant.

## Results

### Characteristics of Study Sample

Table 1 shows the characteristics of our study sample of 50 adults and 30 teenagers. For adults, the mean age was 50.7 years and mean years since diagnosis was 11.7. The sample of adults was mostly female (84.0%), white (94.0%), with 68% holding college or advanced degrees. The majority resided in New Jersey (NJ) (38.0%) or New York City (NYC) (34.0%) and had gastroenterologists affiliated with the Celiac Center at Columbia University (82.0%). Only 16% were currently seeing a RDN. For teenagers, the mean age was 15.7 and mean years since diagnosis was 6.0. The sample of teenagers was also mostly female (80%) and white (96.7%). The majority (73.3%) were still in high school.

**Table 1** Demographic and patient characteristics of study sample

	Adults ( $n = 50$ )	Teenagers ( $n = 30$ )
Age, mean (SD)	50.7 (17.8)	15.7 (1.5)
BMI, mean (SD)	23.3 (3.4)	20.0 (2.4)
Years since diagnosis, mean (SD)	11.7 (10.8)	6.0 (4.6)
1–4	8 (16.3)	14 (46.7)
5–10	21 (42.9)	10 (33.3)
> 10	20 (40.8)	6 (20.0)
Female gender ( $n, \%$ )	42 (84.0)	24 (80.0)
Race ( $n, \%$ )		
White	47 (94.0)	29 (96.7)
Other	3 (6.0)	1 (3.3)
Education		
Middle school	–	7 (23.3)
High school	4 (8.0)	22 (73.3)
Some college	12 (24.0)	1 (3.3)
College graduate	14 (28.0)	
Postgraduate	20 (40.0)	
Residence (based on zip code) ( $n, \%$ )		
NJ	19 (38.0)	10 (33.3)
NYC	17 (34.0)	8 (26.7)
Westchester	9 (18.0)	7 (23.3)
Others (NYS, CT LI)	5 (10.0)	5 (16.7)
Gastroenterologist affiliated with Celiac Center	41 (82.0)	23 (76.7)
Visits with RDN		
RDN currently	8 (16.0)	8 (26.7)
RDN past only (once)	25 (50.0)	11 (36.7)
RDN past only (more than once)	12 (24.0)	2 (6.7)
RDN never	5 (10.0)	9 (30.0)

BMI body mass index, NJ New Jersey, NYC New York City, NYS New York State, CT Connecticut, LI Long Island, RDN Registered Dietitian Nutritionist

Most resided in NJ (33.3%) or NYC (26.7%) and had gastroenterologists affiliated with the Celiac Center of Columbia University (76.7%). A little over one-quarter were currently seeing an RDN.

### Differences in CD-Specific QOL, Knowledge, and Adherence by Energy Level

Table 2 shows differences in CD-specific QOL, knowledge, and adherence by energy level. For adults, the overall mean (SD) CDQOL score was 74.1 (14.2) which corresponds to a good QOL [23]. Adults with lower energy levels had significantly lower overall CD-specific QOL scores than adults with higher energy levels [68.0 (13.6) vs 78.9 (13.0),  $p = 0.006$ ]. A similar pattern was seen for 2 of the 4 subscales. Thus, for adults, lower energy level was associated with more dysphoria ( $p = 0.024$ ) and more reported limitations ( $p = 0.002$ ). Adults with lower energy level also displayed greater knowledge (5.6 (0.6) vs 5.0 (0.9),  $p = 0.012$ ).

For teenagers, the overall mean (SD) CDPQOL score was 70.1 (14.9) which also corresponds to a good QOL [24]. Those with lower energy levels had significantly lower overall CDPQOL scores than teens with higher energy levels (59.2 (16.7) vs 73.5 (13.0),  $p = 0.024$ ). A similar pattern was seen for 2 of the 4 subscales (social and isolation). Thus, for teenagers, lower energy levels were associated with more social concerns ( $p = 0.026$ ) and greater feelings of isolation

( $p = 0.017$ ). Teenagers with lower energy levels also displayed greater knowledge (5.4 (0.5) vs 4.7 (0.9),  $p = 0.057$ ), although this association was not statistically significant.

### Differences in CD-Specific QOL and Knowledge by Dietary Vigilance Level

Table 3 shows differences in CD-specific QOL and knowledge by level of dietary vigilance. Extremely vigilant adults ( $n = 12$ ) had significantly lower overall QOL scores and subscales than adults who were less vigilant [64.2 (16.0) vs 77.2 (12.2),  $p = 0.004$ ]. Adults that were extremely vigilant displayed more dysphoria ( $p = 0.016$ ), more reported limitations ( $p = 0.004$ ), and more concerns about inadequate treatment ( $p = 0.012$ ). At the same time, extreme vigilance was also associated with greater knowledge [5.7 (0.7) vs 5.1 (0.8),  $p = 0.035$ ]. The same patterns were not evident among teenagers among whom there were no significant differences by level of dietary vigilance.

### Barriers and Facilitators

Table 4 summarizes major themes that arose from the open-ended barrier and facilitator questions about maintaining a strict gluten-free diet. Sample responses for each theme are shown in Tables 5 (barriers) and 6 (facilitators). Barrier responses fell into two main categories: those related

**Table 2** Celiac disease-specific quality of life (QOL)<sup>a</sup>, knowledge<sup>b</sup>, and adherence<sup>c</sup> in adults and teenagers by energy level

Adults	Higher energy ( $n = 28$ )	Lower energy ( $n = 22$ )	Total ( $n = 50$ )	<i>t</i>	<i>p</i>
Overall CD-QOL (0–100)	78.9 (13.0)	68.0 (13.6)	74.1 (14.2)	2.9	0.006
Subscales					
Dysphoria	95.1 (9.1)	88.1 (12.2)	92.0 (11.0)	2.3	0.024
Limitations	76.4 (16.5)	61.1 (16.8)	69.7 (18.2)	3.2	0.002
Health concerns	71.8 (18.7)	66.8 (20.7)	69.6 (19.5)	0.9	0.377
Inadequate treatment	74.6 (22.7)	61.9 (27.9)	69.0 (25.7)	1.8	0.084
Knowledge	5.0 (0.9)	5.6 (0.6)	5.2 (0.8)	−2.6	0.012
Adherence (SDE)	1.4 (0.5)	1.2 (0.2)	1.3 (0.4)	2.0	0.056
Teenagers	Higher energy ( $n = 23$ )	Lower energy ( $n = 7$ )	Total ( $n = 30$ )	<i>t</i>	<i>p</i>
Overall CDPQOL	73.5 (13.0)	59.2 (16.7)	70.1 (14.9)	2.4	0.024
Subscales					
Social	73.6 (15.1)	58.2 (15.1)	70.0 (16.3)	2.4	0.026
Uncertainty	69.6 (21.1)	67.9 (12.2)	69.2 (19.2)	0.2	0.841
Isolation	78.5 (12.9)	59.8 (27.4)	74.2 (18.6)	2.5	0.017
Limitations	70.3 (21.3)	52.4 (29.5)	66.1 (24.2)	1.8	0.086
Knowledge	4.7 (0.9)	5.4 (0.5)	4.9 (0.9)	−2.0	0.057
Adherence (SDE)	1.3 (0.3)	1.3 (0.2)	1.3 (0.3)	0.0	0.988

<sup>a</sup>Higher CDPQOL (adults) or CDPQOL (teenagers) scores and subscales suggest higher degree of QOL; Scales 0–100

<sup>b</sup>Higher knowledge scores suggest higher knowledge; Scale 0–6

<sup>c</sup>Higher adherence scores suggest lower adherence; Scale 1–6

**Table 3** Celiac disease-specific quality of life (QOL)<sup>a</sup> and knowledge<sup>b</sup> in adults and teenagers by vigilance level

Adults	Extremely vigilant ( <i>n</i> = 12)	Less vigilant ( <i>n</i> = 38)	Total ( <i>n</i> = 50)	<i>t</i>	<i>p</i>
Overall CDQOL	64.2 (16.0)	77.2 (12.2)	74.1 (14.2)	− 3.0	0.004
Subscales	85.4 (15.8)	94.1 (8.2)	92.0 (11.0)	− 2.5	0.016
Dysphoria	56.7 (18.2)	73.8 (16.3)	69.7 (18.2)	− 3.1	0.004
Limitations	65.0 (20.4)	71.1 (19.3)	69.6 (19.5)	− 0.9	0.36
Health concerns	53.1 (25.1)	74.0 (24.0)	69.0 (25.7)	− 2.6	0.012
Inadequate treatment knowledge	5.7 (0.7)	5.1 (0.8)	5.2 (0.8)	2.2	0.035
Teenagers	Extremely vigilant ( <i>n</i> = 7)	Less vigilant ( <i>n</i> = 23)	Total ( <i>n</i> = 30)	<i>t</i>	<i>p</i>
Overall CDOPQOL	74.6 (14.2)	68.8 (15.2)	70.1 (14.9)	0.9	0.38
Subscales	72.5 (16.7)	69.3 (16.5)	70.0 (16.3)	0.4	0.66
Social	66.7 (27.6)	69.9 (16.6)	69.2 (19.2)	− 0.4	0.70
Uncertainty	83.9 (7.1)	71.2 (20.1)	74.2 (18.6)	1.6	0.12
Isolation	75.0 (15.2)	63.4 (26.0)	66.1 (24.2)	1.1	0.27
Limitations knowledge	5.1 (0.7)	4.8 (0.9)	4.9 (0.9)	0.9	0.39

<sup>a</sup>Higher CD-QOL (adults) or CDPQOL (teenagers) overall scores and subscales suggest higher degree of QOL; Scales 0–100

<sup>b</sup>Higher knowledge scores suggest higher knowledge; Scale 0–6

to eating out and those related to gluten-free products (or the lack thereof). For adults, the main barriers to eating out included constant worries about cross-contamination (34%), distrust of the gluten-free menu designation (30%), dislike of having to constantly ask questions related to their diet and/or advocate for safe food (30%), limited restaurant choices (24%) dismissive or uninformed wait staff (22.0%), and the general perception that gluten free is a fad or weight loss diet (16%). Themes related to gluten-free products included missing specific foods like pizza, bread, or pasta (38%), the added expense of gluten-free options (22%), and the need for better label laws for both food (16%) and pharmaceuticals and makeup (16%). Less common, but described, were concerns about gluten-free foods being unhealthy (12%) and unpalatable (10%). There were no significant differences between the 12 “extremely vigilant” adults and their less vigilant counterparts, but, for nine of twelve barrier themes, percentages were higher among the extremely vigilant. The distribution of barrier themes was similar for teenagers, although adults were more likely to mention lack of label laws (16.0 vs 0.0%,  $\chi^2 = 3.7$ , *df* = 1, *p* = .054).

For adults, the main facilitator themes that emerged were having supportive friends and family (70%), having more gluten-free product options (52%), cooking at home versus eating out (48%), having more gluten-free restaurant options (36%), increased general awareness of the public about CD (26%), and having helpful apps and internet sites (20%). Less common, but described, were facilitators related to having improved label laws (14%), having resources from the Celiac Disease Center or from support groups (10%), and having other family members with CD that related to their

needs (4%). The 12 “extremely vigilant” adults were more likely than their less vigilant peers to mention helpful apps/internet sites (50.0 vs 10.5%,  $\chi^2 = 6.6$ , *df* = 1, *p* = .010) and were nearly significantly more likely to mention cooking at home versus eating out (75.5 vs 39.5%,  $\chi^2 = 3.3$ , *df* = 1, *p* = .069). For nine of ten facilitator themes, percentages were higher among the “extremely vigilant” adults. The distribution of facilitator themes was similar for teens. Adults were nearly significantly less likely to mention supportive family/friends (70.0 vs 90.0%,  $\chi^2 = 3.2$ , *df* = 1, *p* = .072).

In the context of being asked about barriers, 56% of adults and 70% of teens explicitly referenced the adverse social impact of adhering to a gluten-free diet. Words that came up included “misunderstood,” “embarrassed,” “different,” “stigmatized,” “left out,” “awkward,” “guilty.” Some suspected they were not invited to events or homes because of their dietary restrictions. Others dreaded having to explain their situation to new friends. There was resentment for the fact that they could not be “spontaneous” like their peers and that they often had to bring their own lunch or snacks. Three teens expressed apprehension at having to manage their own diet when they went away to college.

## Discussion

This study is the first we are aware of that highlights the potential negative consequences of hypervigilance to a strict gluten-free diet for individuals with CD. The extremely vigilant adults in our sample (i.e., those who consistently ate at celiac-friendly restaurants, asked thorough questions when

**Table 4** Percentage of respondents mentioning main barrier and facilitator themes for adhering to strict gluten-free (GF) diet (based on qualitative data) for adults and teens: comparison of extremely vigilant (EV) versus less vigilant (LV) respondents

	Adults			Teens		
	EV <sup>a</sup>	LV <sup>a</sup>	Total	EV <sup>a</sup>	LV <sup>a</sup>	Total
<i>Barriers</i>						
<i>Eating out</i>						
Risk of cross-contamination	41.7	31.6	34.0	28.6	43.5	40.0
Untrustworthy GF menus	33.3	28.9	30.0	28.6	17.4	20.0
Need to ask questions/prepare ahead/advocate	33.3	28.9	30.0	42.9	30.4	33.3
Limited restaurant choices	16.7	26.3	24.0	42.9	26.1	30.0
Dismissive/uninformed wait staff	33.3	18.4	22.0	0.0	13.0	10.0
Faddishness undermining seriousness	16.7	15.8	16.0	28.6	8.7	13.3
Percentage reporting $\geq 1$ eating out theme	83.3	71.0	74.0	85.7	87.0	86.7
<i>GF products</i>						
Missing specific foods (e.g., pizza, bread, pasta) <sup>a</sup>	41.7	36.8	38.0	57.1	39.1	43.3
Added expense of GF foods	16.7	23.7	22.0	14.3	17.4	16.7
Lack of label laws <sup>b</sup>	16.7	15.8	16.0	0.0	0.0	0.0
Determining gluten in pharmaceuticals/makeup	25.0	13.2	16.0	0.0	4.3	3.3
Unhealthy GF foods	16.7	10.5	12.0	0.0	0.0	0.0
Unpalatable GF foods	0.0	13.2	10.0	14.3	13.0	13.3
Percentage reporting $\geq 1$ GF products theme	75.0	71.1	72.0	71.4	47.8	53.3
<i>Facilitators</i>						
Supportive family/friends <sup>b</sup>	83.3	65.8	70.0	85.7	91.3	90.0
Accessibility of GF products	50.0	52.6	52.0	71.4	43.5	50.0
Cooking at home versus eating out	75.0	39.5	48.0 <sup>c</sup>	71.4	43.5	50.0
Accessibility of GF restaurant options	33.3	36.8	36.0	14.3	56.5	46.7
Increased general knowledge/awareness	41.7	21.1	26.0	28.6	4.3	10.0
Helpful apps/internet sites	50.0	10.5	20.0 <sup>c</sup>	42.9	21.7	26.7
Improved labeling	25.0	10.5	14.0	0.0	4.3	3.3
Celiac Center or support groups	8.3	10.5	10.0	0.0	4.3	3.3
Having other family members with celiac disease	0.0	5.3	4.0	14.3	13.0	13.3
Percentage reporting $\geq 1$ facilitators	100.0	100.0	100.0	100.0	100.0	100.0

<sup>a</sup>Adults: pizza (8%), fast food options (16%), bread (10%), beer/vodka (10%), pasta (6%), dessert (4%), breakfast options (6%), Chinese food (10%), Italian food (2%), teens: pizza (13%), snacks (13%), bread (7%), pasta (7%), dessert (7%), breakfast options (3%), Chinese food (3%), Italian food (3%)

<sup>b</sup>Adult versus teen total  $p < .10$ : lack of label laws  $\chi^2 = 3.7$ ,  $df = 1$ ,  $p = .054$ ; supportive family/friends  $\chi^2 = 3.2$ ,  $df = 1$ ,  $p = .072$

<sup>c</sup>EV versus LV  $p < .10$ : cooking at home versus eating out  $\chi^2 = 3.3$ ,  $df = 1$ ,  $p = .069$ ; helpful apps/internet sites  $\chi^2 = 6.6$ ,  $df = 1$ ,  $p = .010$

dining out, eliminated cross-contamination potential in their home kitchen.) had significantly lower QOL scores than their less vigilant counterparts. Our qualitative data suggested that cooking at home (as opposed to eating out) and using internet sites and apps to facilitate gluten avoidance were particularly prevalent strategies used by the hypervigilant to maintain a strict gluten-free diet. The qualitative data also highlighted the particulars that make eating out problematic for individuals with CD.

Our findings are in contrast with several others that found better dietary adherence to be associated with higher QOL scores [18–21] or that found no association [27, 28]. Inconsistent findings may, in part, be due to differences in quality

of life instruments (generic vs CD specific), differences in dietary adherence instruments (self-report vs RDN or health professional opinion base on the dietary data), or geographic location. In this study, we used validated celiac-specific quality of life measures (CDQOL and CDPQOL) and relied on nutrition professionals' opinions about participant's vigilance based on interviews and 24-h recall data (as opposed to self-reported perceptions of subject's own dietary adherence). For example, one participant perceived herself to be extremely vigilant (i.e., reporting that she "never" had gluten over the past month), but her interview and 24-h recalls revealed a major source of cross-contamination (e.g., pulling the croutons off of her Caesar salad before eating it). By

**Table 5** Sample responses for barriers to adhering to strict gluten-free (GF) diet (based on qualitative data)

Barrier	
Risk of cross-contamination	The salad bar was marked as GF but was near croutons and stuff—it mixes! Salad with side of bread or ice cream with cookies can come automatically. Will ask to send back unless obvious the foods did not come in contact
Untrustworthy gluten-free menus	The menu said GF and I got sick most times I was there Many menus will say they are GF but clearly they are not (e.g., a salad that has pita bread, dressing that's not GF)
Need to ask questions/prepare ahead/advocate	You have to constantly be aware, especially in public settings. It feels like you are constantly advocating. Having to be prepared all the time carrying snacks, checking restaurants ahead of time, etc. Keeping other people educated can be exhausting. There is a lot of anxiety going to a restaurant –being in line asking questions—I don't want to hold up the line
Limited restaurant choices	I wish places like Duncan Donuts or Starbucks were more considerate and have gluten free snacks; big chains should have more options I feel like I have to go to same restaurants all the time that I know are safe
Dismissive or uninformed wait staff	For example, I was at pizza restaurant and asked about the fryer. I learned it was used for gluten-containing foods so I didn't order anything fried. Instead, I ordered the GF broccoli pizza and later realized the broccoli was fried. I was upset that the staff wasn't knowledgeable enough to know I shouldn't have had broccoli on the pizza despite having asked a lot of questions
Faddishness undermining seriousness	Restaurants complain that they are providing GF foods to people who don't seem to be consistent about needing to eat GF Going out to eat is a problem especially because it's a fad diet and not everyone takes it seriously
Missing specific foods (e.g., pizza, bread, pasta)	I can't find good bread! All are small, bad texture There are some commercial products like goldfish and ice cream cakes that don't yet have a GF option or the GF options don't approximate it
Added expense of gluten-free foods	My food costs have doubled The cost of gluten free flours for baking are expensive The restaurants that are safe are very expensive
Lack of label laws	It is frustrating that in the US it is often difficult to know if foods/products contain gluten. In South Africa everything is labeled containing gluten/not containing gluten. Ireland also labels everything
Safety of pharmaceuticals or makeup	It's hard finding soaps and shampoos that are GF. In public restrooms, I don't like to use the soap—I'm never sure if should bring own
Unhealthy gluten-free foods	Most gluten free products are junk food and poor diet quality It is difficult to get adequate nutrients on a GF diet, specifically fiber and whole grains
Unpalatable gluten-free foods	Sometimes I don't tell the restaurant I am GF or I get steamed/boiled chicken that is really bad

classifying participants' vigilance level based on nutrition professional's opinion, we can more confidently assert that the "extremely vigilant" group were truly those taking the greatest precautions to avoid all sources of gluten.

Our data provide insights into the ways individuals with CD struggle with eating out. Seventy-four percent of adults and 86.7% of teens mentioned one or more barriers to adhering to a gluten-free diet that were related to eating out. The increasing popularity of the gluten-free diet in non-celiac individuals [29] in the U.S. has created a variety of challenges for those with CD. The seriousness of gluten exposure may now be dismissed at restaurants for being "trendy," forcing CD individuals to advocate even harder. Our findings are in contrast to those conducted in Europe where the gluten-free diet is recognized, even in restaurants, as a medical necessity for CD [18, 27]. Furthermore, while increasing numbers of restaurants offer gluten-free menu options, it

is unclear what steps are taken to ensure that food service staff handles food properly and with regular monitoring and quality control. We found the desire to take advantage of increased restaurant options, combined with the distrust of menus and ill-informed wait staff, to be a source of considerable frustration and anxiety for our participants. For those who were extremely vigilant, the frustration is reflected in their reliance upon cooking at home as opposed to eating out.

Our qualitative data also provide insights into prevalent facilitators to staying gluten free. Supportive family and friends, increased accessibility of gluten-free products, apps and internet sites, and the benefits of cooking were particularly appreciated. Adults who were extremely vigilant particularly relied upon cooking at home versus eating out, and helpful apps and internet sites, more so than their less vigilant counterparts. We believe identification



**Table 6** Sample responses to facilitators to adhering to strict gluten-free (GF) diet (based on qualitative data)

Facilitators	
Supportive family and friends	My family is a big help. We are strictly GF at home—no one is allowed to bring anything gluten into the home My boyfriend has switched to a mostly GF diet My mom calls places ahead to see what I can eat—she'll even go in a restaurant kitchen! My mom is highly skilled at questioning the waiter!
Accessibility of GF products	GF pretzels—years ago they didn't exist! There are just more products now, sections in food stores are larger, the quality and taste are better
Cooking at home versus eating out	I cook so I don't have to worry I cook which makes it easier I cook a lot so I have a good sense of what ingredients to ask about at restaurants (e.g., chicken broth) I have always cooked so I just cook differently now and then I know it's safe
Accessibility of GF restaurant options	Now you can go anywhere and eat out—you can get steak, chicken, GF pasta and pizza. It's gotten a lot easier over the years
Increased general knowledge and awareness	Being GF is more known now in big cities. More mainstream. More available Being GF is more common now so people make the connection More restaurants are familiar, especially when traveling
Helpful apps and internet sites	FindMeGlutenFree Scanning bar codes
Improved labeling	At least labels now say if a product contains wheat Labeling laws now list the common allergies
Celiac Center or support groups	Meeting with nutritionists at the Celiac Center and at school provides support My support group, which is no longer together, was helpful adapting to diet. I am very grateful for the network/community
Having other family members with CD	Most of my family is Celiac so even at extended family gatherings there are a lot of options. I have a cousin with celiac so that makes it more acceptance at family functions. My aunt makes two turkeys on Thanksgiving

of these facilitators can inform future educational efforts to increase adherence to a gluten-free diet.

Our results suggest the importance of ongoing involvement of a registered dietitian nutritionist (RDN) with celiac patients, involvement that persists beyond the initial diagnosis [6, 7]. Conversations to promote dietary adherence and ensure a high quality of life will take time and cannot be done in a single visit. Our data, as well as the literature, suggest that regular dietitian follow-up falls short of guidelines. Of particular concern was that only 16% of adults and 26.7% of teenagers in our sample were currently seeing a RDN. Most had a visit with a RDN when first diagnosed, but without follow-up. Several had never had a visit with a RDN. Our participants reported relying on the internet for guidance, despite the fact that most popular CD websites have been shown to be insufficiently accurate, comprehensive, or trustworthy [30]. Lack of insurance and/or limited availability of dietitians specialized in CD were not likely to be problems for our study population. The NIH consensus development conference on CD advised that patients undergo consultation with a skilled RDN and continuous long-term follow-up by a multidisciplinary team. It appears that these guidelines are rarely met among our participants who were contacted via a celiac disease center email list [31].

From a clinician's perspective, we believe our findings have clinical relevance. In our study, adults who were extremely vigilant versus less vigilant differed by at least 10 points on the overall CDQOL scale, as well as the dysphoria CDQOL subscale. And those who were extremely vigilant versus less vigilant differed by approximately 20 points on the limitations and inadequate treatment CDQOL subscales. When the CD-QOL instrument was validated, a difference of approximately 10 points lower on the CD-QOL scale was enough to move individuals into a worse category of self-rated health, psychological distress, functional status, or pain. For example, differences of 10 points on the CD-QOL scale differentiated those that had low versus mid-level psychological distress; differences of 20 points on the CD-QOL scale differentiated those that had low versus high-level psychological distress. Similar patterns were found for the others scales. Thus, the hypervigilance described in our sample (e.g., bringing their own dishes to restaurants or other homes; thorough and repeated questioning at restaurants) may come with a meaningful and relevant cost.

This study has several limitations. First, it was conducted at a single CD referral center, the sample sizes were small and the population was demographically homogeneous. Second, the design was cross sectional and not longitudinal. While we found that "extreme vigilance" (in adults only)

and low energy (in adults and teens) were associated with lower CD-specific QOL, we cannot say if “extreme vigilance” causes lower energy and QOL or if lower QOL and energy lead to extreme vigilance. We speculate that, in some individuals, extreme vigilance to a strict gluten-free diet may be creating anxiety and stress leading to a lower QOL. This anxiety and stress, may, in turn, lead to lack of sleep and even depression, which may lead to low energy levels or fatigue. The relationships need to be explored prospectively to determine the direction of causality. Third, our measure of energy level was limited to a single item on the CDAT. A more extensive questionnaire on symptoms would have been preferable. Fourth, vigilance was based on 24-h recalls which rely on participant’s memory of what they ate and the precautions they took to avoid gluten. Thus, there was the potential for misclassification on level of vigilance. Finally, our exploration of barriers to and facilitators of maintaining a gluten-free diet were based on open-ended questions. Themes were overlapping and it is likely that different people were expressing the same idea, but with different choices of words. When considering the percentages reporting each theme, it must be kept in mind that study subjects were not given the opportunity to agree or disagree with each statement.

Our data illustrate the critical need to develop and evaluate nutrition education strategies that promote increased adherence to a gluten-free diet while, *at the same time*, taking care to maintain high QOL. Given the seriousness of poor adherence and psychological burden, it is surprising that there is so little research conducted on alternative approaches to improve gluten-free diet adherence and QOL. In fact, only five behavioral intervention studies have been reported with CD patients [32–36] of which only one [35] targeted adherence and two targeted QOL [34, 36]. Possible future directions would be to explore interventions that combine visits to a RDN (i.e., standard of care) with strategies designed to address barriers associated with eating out (e.g., portable gluten sensor monitoring or promotion of skills to combat the perception that gluten free is only a fad). Interventions that promote cooking skills could decrease reliance on eating out. Since over 80% of participants reported the importance of supportive friends and family, it is important to determine the impact of family-centered nutrition education when promoting dietary adherence. Ultimately, we need longitudinal studies to test the best level of dietary adherence that can avoid symptoms, intestinal damage, and long-term complications, yet maximize energy levels and quality of life for the celiac population. We also need to determine the significance of potential sources of cross-contamination for the risk of ingesting small quantities of gluten. Until then, we must advocate for a strict gluten-free diet with the caveat that, for some, such hypervigilance comes at a cost that needs to be supported and addressed.

## Conclusion

In this prospective, cross-sectional study, we identified potential negative consequences of hypervigilance to a strict gluten-free diet for individuals with CD. Clinicians need to be aware of the importance of promoting both dietary adherence and quality of life. While patients must be encouraged to continue following a strict 100% gluten-free diet, we hope that our findings highlight the importance of clinicians addressing both adherence to a strict gluten-free diet while concurrently addressing emotional and social well-being while caring for their patients with celiac disease. Our findings suggest that there may be a cost to such hypervigilance and interventions that promote *both* strict adherence and maximize quality of life are urgently needed.

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**Author’s contribution** RW, BL, AL, NR, and PG conceptualized and designed the study. JC and CA collected data and contributed to conceptualization of the qualitative analyses. PZ managed and analyzed the data. All authors (RW, BL, AL, PZ, NR, JC, CA, PG) reviewed and commented on multiple drafts of the manuscript, and all played a key role in the interpretation of study results.

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## Compliance with ethical standards

**Conflicts of interest** RW, BL, AL, PZ, NR, JC, CA declare that they have no conflicts of interest. PG serves on the Advisory board of ImmusanT, Cellimmune and ImmunogenX.

**Ethical approval** All procedures performed in this study involving human participants were in accordance with ethical standards of the institutional review boards at both Columbia University Medical Center and Teachers College, Columbia University and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

**Informed consent** Written informed consent was obtained from all individual participants included in the study.

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