ADOLESCENTS AND YOUNG ADULTS

Diminished quality of life among adolescents with coeliac disease using maladaptive eating behaviours to manage a gluten-free diet: a cross-sectional, mixed-methods study

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Abstract

Background: Certain approaches to managing a strict gluten-free diet (GFD) for coeliac disease (CD) may lead to impaired psychosocial well-being, a diminished quality of life (QOL) and disordered eating. The present study aimed to understand adolescents' approaches to managing a GFD and the association with QOL.

Methods: Thirty adolescents with CD (13–17 years old) following the GFD for at least 1 year completed the Celiac Dietary Adherence Test (CDAT) and QOL survey. Their approaches to GFD management were explored using a semi-structured interview, where key themes were developed using an iterative process, and further analysed using a psychosocial rubric to classify management strategies and QOL. CDAT ratings were compared across groups.

Results: Gluten-free diet management strategies were classified on a four-point scale. Adaptive eating behaviours were characterised by greater flexibility (versus rigidity), trust (versus avoidance), confidence (versus controlling behaviour) and awareness (versus preoccupation) with respect to maintaining a GFD. Approximately half the sample (53.3%) expressed more maladaptive approaches to maintaining a GFD and those who did so were older with lower CD-Specific Pediatric Quality of Life (CDPQOL) scores, mean subscale differences ranging from 15.0 points for Isolation ($t = 2.4$, $P = 0.03$, d.f. = 28) to 23.4 points for Limitations ($t = 3.0$, $P = 0.01$, d.f. = 28).

Conclusions: Adolescents with CD who manage a GFD with maladaptive eating behaviours similar to known risk factors for feeding and eating disorders experience diminished QOL. In accordance with CD management recommendations, we recommend ongoing follow-up with gastroenterologists and dietitians and psychosocial support referrals, as needed.

Introduction

Coeliac disease (CD) is a multisystem autoimmune disorder that damages the small intestine and is triggered by dietary gluten, the main protein found in wheat, barley, and rye [1]. CD affects approximately 1% of the US population [2]. Current treatment for CD requires strict avoidance of all foods that contain gluten [3]. For those with CD, exposure to even a small amount of gluten may cause a range of symptoms [3–5] and complications

including anaemia, osteoporosis and cancer [6,7]. Observational studies using duodenal mucosal healing as a marker of adherence suggest that adherence to a strict gluten-free diet (GFD) can help to decrease symptoms and the risk of long-term complications [8,9].

Maintaining the GFD requires increased control around food, including monitoring of food labels, avoiding unsafe grains and detailed questioning to avoid cross-contact with gluten containing foods. These constraints can be hard to accept and follow, especially for adolescents, comprising an age group typically burdened by acute concern regarding fitting in with peers, wanting to feel normal and gluten-free food displeasure [10–14]. Adherence and self-advocacy are particularly problematic for adolescents as they venture outside of the home and become more independent [13,14]. The focus on food and eating behaviours, and possibly even adherence to a GFD, may be associated with an increased risk of psychosocial problems and disordered eating patterns and behaviours in adolescents with CD [15–21].

A spectrum of behavioural health difficulties exists of feeding and eating disorders [21]. Disordered eating behaviours include dieting, purging, binge eating, fasting and the use of excessive physical activity to control weight and/or body shape [21–24]. Although healthy eating patterns fluctuate based on factors such as food availability and proximity, fluctuation to the point of nutrient deficiency or excess weight change, indicate disordered eating [21–25]. Planning and preparing food should not dominate thoughts and dictate behaviours above and beyond that of other daily activities [24]. Recent research on disordered eating in CD indicates that both extreme GFD adherence and dietary transgressions are associated with disordered eating patterns [26–32]. For those who fail to adhere to their GFD, the challenges of managing the GFD may lead to restrictive or bulimic eating behaviours [31–33]. For those who are extremely adherent to the GFD, anxiety around gluten cross-contamination may lead to limited food choices or eating only in situations with complete control over food preparation, which may mimic disordered eating [21,27,34,35]. Patterns of cognition and behaviour that cause individuals to become socially isolated, to refuse attendance at social events involving food or to avoid eating in settings outside the house, a marked interference with psychosocial functioning, can be considered disordered [22]. Currently, there is limited description of how individuals adhere to the GFD.

In a previous study, we found that hypervigilance to a strict GFD among adults with CD correlated with a diminished quality of life (QOL); the relationship was present but less robust among adolescents [36]. In the present study, our objective was to analyse data from a semi-structured interview describing adolescent approaches to managing a strict GFD, as well as to identify associations between these approaches and CD-Specific Pediatric Quality of Life (CDPQOL). The interview explored barriers and facilitators to adhering to a strict GFD. An inductive content analytic approach was used to identify management approaches which might affect QOL. These data will help to inform future nutrition education strategies which can promote a strict GFD at the same time as maximising QOL for adolescents with CD.

Materials and methods

Recruitment

Data were collected in the course of a cross-sectional prospective study of both adults ($n = 50$) and adolescents ($n = 30$) with CD [36]. The present analysis is restricted to the adolescent subgroup (13–17 years old). The study was conducted at the Celiac Disease Center in New York City at the Columbia University Medical Center. Adolescents, who were no older than 17 years at enrollment, with a self-reported duodenal biopsy-confirmed diagnosis of CD, in accordance with US guidelines [37] at least 1 year prior to enrollment and willing to participate in three visits (the first one in-person and two subsequent ones by telephone) over a 1-month period were included. Exclusions included: (i) CD diagnosis <1 year prior to enrollment; (ii) serum or self-diagnosed CD (without biopsy); and (iii) age <13 years old. Completers received a $25 Amazon gift card.

Enrollment was between March and August 2016. Our target enrollment was 30 adolescents. Those affiliated with the Celiac Disease Center of Columbia University (approximately 5000 members: those interested in CD, including a mix of patients and family members) were e-mailed to ascertain interest in the study. There were two additional follow-up emails. Among the 45 adolescents who responded to the email invitation, 15 were ineligible (11 lacked duodenal biopsy to confirm CD; three never scheduled an appointment) and 30 were eligible and enrolled.

Demographic and medical history variables

Gender (male, female), age, self-described race (White, African-American, Asian, Other), self-described ethnicity (Hispanic, non-Hispanic), zip code of home residence and education (highest level/grade achieved) were assessed [36]. Medical history included self-reported height, weight and years since CD diagnosis.

CD-Specific Pediatric Quality of Life

CDPQOL was evaluated with a 17-item validated survey instrument [38]. Participants responded to Likert scales

questions ranging from 0 = Never to 4 = Almost Always. Answers were transformed and combined, obtaining four subscales and a Total score. Social items (n = 7) measured how much respondents feel misunderstood, are a burden and their self-esteem. Uncertainty items (n = 3) measured how much they worry about their future, college and getting older with CD. Isolation items (n = 4) measured how much participants feel different from their friends and family because of their CD diagnosis. Limitation items (n = 3) measured how much they feel nervous about eating at friends' homes or avoid parties. Higher CDPQOL scores indicated better QOL, where the possible overall score range was 0–100.

Gluten-free diet adherence and symptoms

Dietary adherence was measured using the Celiac Dietary Adherence Test (CDAT) [39]. The CDAT is a seven-item validated, self-administered, survey instrument that includes two items about persistent symptoms (i.e. headaches and low energy) and five items about attitudes and behaviours related to gluten exposure (including one item that specifically asks about frequency of eating gluten on purpose). Total scores range from 7 to 35, with higher scores implying worse adherence to the GFD. Total scores >13 indicate poor adherence [39]. The symptoms subscale ranges from 2 to 10, with a higher score implying worse symptoms. Eating gluten on purpose scores ranged from 1 to 5, with higher scores implying worse adherence to the GFD. Three 24-h dietary recalls were also collected over a 1-month period and reviewed for quantity and frequency of intentional or unintentional gluten exposure by determining level of vigilance [36] (e.g. asks thorough questions when dining out, has eliminated cross-contamination potential in kitchen, review of diet reveals no obvious gluten sources, etc.) each recorded on a six-point Likert scale ranging from 1 (excellent adherence) to 6 (not following a GFD). Those 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).

Adaptive and maladaptive eating patterns

Because no tools are available to assess eating patterns in CD, a semi-structured interview was used to explore key themes concerning the management of the GFD. Adolescents were asked: (i) What do you see as the major challenges to following a strict GFD? (ii) What do you see as the things that help make it easy to follow a strict GFD? Prompts to elicit clarity were offered, including to

understand motivations, as appropriate. At the end of each interview, adolescents were given the opportunity to discuss other issues, apart from their parents, including intentional gluten consumption. Interviewers hand transcribed detailed notes during the interview. Interview responses formed the basis of our classification of eating behaviours as described below.

Statistical analysis

Analysis of quantitative data

Frequencies and percentages are presented for categorical variables, means and SDs for continuous data. Differences by gender and percentage Extremely Vigilant between eating behaviour groups were assessed with chi-squared tests. Differences by continuous variables (i.e. age, body mass index (BMI), CDPQOL and CDAT) were assessed with one-way analyses of variance and t-tests. As appropriate, comparisons of continuous variables were repeated using analysis of covariance (ANCOVA) to control for age at enrollment. SPSS, version 19.0 (IBM Corp., Armonk, NY, USA) was used for the analysis.

Qualitative analysis of adaptive and maladaptive eating classification scheme

Consistent with Fade and Swift [40], responses to the semi-structured interviews were read repeatedly by the first author and inductively coded on paper copies first literally, and then into interpretative themes. Key themes were developed into a framework for coding the entire dataset. To enhance reliability, the coding process and emerging themes were discussed among the authors until consensus was achieved. A decision trail was used to ensure transparency.

The analysis of the semi-structured interview data was guided by a psychosocial rubric previously used to identify management strategies and disordered eating in patients with diabetes [41] (see Supporting information, Appendix S1). Individuals were classified as having adaptive or maladaptive eating thoughts and behaviours. Adaptive eating behaviours were characterised by greater flexibility, trust, confidence and a less active focus on maintaining a GFD (i.e. acceptance versus preoccupation). Maladaptive eating behaviours were characterised by greater rigidity, avoidance, controlling behaviour and preoccupation with maintaining a GFD.

Participants were classified into four groups by two researchers who were blinded to the CDPQOL scores. Group 1 described only adaptive eating behaviours; Group 2 those who expressed mostly adaptive behaviours, with a few maladaptive behaviours; Group 3 expressed several maladaptive behaviours, with few adaptive behaviours; and Group 4 had mostly maladaptive behaviours. One of the interviewers had participated in the original interviews.

Discrepancies between the two coders were resolved by discussion and review of 24-h dietary recall, if needed.

Ethical approval

The Institutional Review Boards at both Teachers College, Columbia University and the Columbia University Medical Center approved this study. Written consent was obtained from all participants.

Results

Characteristics of study sample by eating behaviour groups
All 30 participants [80% female; mean (SD) age 15.6 (1.5) years] completed the study. Approximately half of adolescents (53.3%, $n = 16$) were classified as having maladaptive eating behaviours (i.e. Groups 3 and 4) (Table 1). Maladaptive eating behaviour was associated with an increased mean (SD) age at study enrollment [16.4 (1.0) years versus 14.7 (1.5) years, $t = -3.6$, d.f. = 1, $P = 0.001$]. Neither age at diagnosis, nor body mass index was associated with maladaptive eating behaviour. There was no significant relationship between gender and eating behaviour group.

CD-Specific Pediatric Quality of Life, symptoms and adherence by eating behaviour groups

Table 2 presents the relationships between CDPQOL, CDAT scores, eating behaviour status and level of strict adherence.

Total CDPQOL score was positively associated with eating behaviour status (linear trend $F = 7.0$, d.f. = 1, $P = 0.01$), as was the Social subscale (linear trend $F = 4.5$, d.f. = 1, $P = 0.04$), Uncertainty subscale (linear trend $F = 4.1$, d.f. = 1, $P = 0.05$) and Limitations subscale (linear

trend $F = 4.5$, d.f. = 1 $P = 0.04$). In each case, maladaptive eating was associated with diminished CDPQOL. Although the linear trend for the Isolation subscale did not attain significance ($F = 2.0$, d.f. = 1, $P = 0.17$), the pattern of means was consistent with that of the other subscales. The two maladaptive eating behaviour groups had a lower Total and CDPQOL subscale scores than the two adaptive eating behaviour groups and the differences in mean scores, ranging from 15.0 for Isolation to 23.4 for Limitations.

Because CDPQOL of scores tended to be negatively correlated with age at enrollment (CDPQOL subscale scores Social: $r = -0.51$, $P = 0.004$, Uncertainty: $r = -0.42$, $P = 0.020$, Isolation: $r = -0.25$ $P = 0.19$, Limitations: $r = -0.31$, $P = 0.10$, Total: $r = -0.49$, $P = 0.006$), the two group comparisons were repeated using ANCOVA. Significant differences remained after controlling for age at enrollment for CDPQOL subscale scores Uncertainty (Main Effects $F = 4.6$, d.f. = 1, $P = 0.04$), Limitations (Main Effects $F = 5.2$, d.f. = 1, $P = 0.03$) and Total (Main Effects $F = 6.4$, d.f. = 1, $P = 0.02$).

The CDAT total score did not differ by eating behaviour group. However, the two-item Headache and Low Energy subscale score trended toward being worse (i.e. higher) in the Maladaptive groups ($t = -1.9$, $P = 0.06$), whereas the single item about eating gluten on purpose was significantly better (i.e. lower) ($t = 2.0$ $P = 0.05$). The maladaptive groups had a higher percentage of the extremely vigilant (31.3% versus 14.3%) but, because there were only seven extremely vigilant participants, the difference was not significant ($\chi^2 = 0.5$, $P = 0.47$).

Qualitative assessment of adaptive and maladaptive eating characteristics

Flexibility versus rigidity
Adolescents with adaptive eating behaviours expressed flexibility in their approach to maintaining a GFD, were able

Table 1 Age and body mass index by eating behaviour

	Most adaptive ($n = 7$) n (%)	Adaptive ($n = 7$) n (%)	Maladaptive ($n = 11$) n (%)	Most maladaptive ($n = 5$) n (%)	Total ($n = 30$) n (%)	Linear trend*		Adaptive versus Maladaptive[†] ($n = 14$ versus 16)	
						χ^2	P	χ^2	P
Female	5 (71.4)	6 (85.7)	8 (72.7)	5 (100.0)	24 (80.0)	0.6	0.43	0.0	0.86
Male	2 (28.6)	1 (14.3)	3 (27.3)	0 (0.0)	6 (20.0)				
	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	Mean (SD)	F	P	t	P
Age at coeliac disease diagnosis (years)	9.4 (4.6)	9.8 (4.3)	8.7 (4.6)	12.6 (6.4)	9.8 (4.8)	0.9	0.35	−0.2	0.88
Age at enrollment (years)	14.7 (1.7)	14.7 (1.3)	16.1 (1.1)	17.0 (0.6)	15.6 (1.5)	13.0	0.001	−3.6	0.001
BMI kg m^{-2}	18.6 (2.8)	19.8 (2.4)	21.0 (1.6)	19.7 (2.8)	20.0 (2.4)	1.1	0.30	−1.6	0.11

*Linear trend χ^2 and F, both d.f. = 1.
[†]Most adaptive + Adaptive versus Maladaptive + Most maladaptive; χ^2 with continuity correction d.f. = 1; t-test d.f. = 28
BMI, body mass index.

Table 2 CDPQOL*, CDAT[†] and vigilance to a gluten-free diet by eating behaviour

	Most adaptive (n = 7) Mean (SD)	Adaptive (n = 7) Mean (SD)	Maladaptive (n = 11) Mean (SD)	Most maladaptive (n = 5) Mean (SD)	Total (n = 30) Mean (SD)	Linear trend[‡]		Adaptive versus Maladaptive (n = 14 versus 16) t-test[§] ANCOVA[¶]			
						F	P	t	P	F	P
CDPQOL											
Total	76.9 (12.5)	81.9 (11.5)	61.4 (12.2)	63.5 (14.8)	70.1 (14.9)	7.0	0.01	3.9	0.001	6.4	0.02
Social	77.0 (15.3)	79.1 (16.4)	63.0 (12.2)	62.9 (19.3)	70.0 (16.3)	4.5	0.04	2.8	0.01	1.9	0.18
Uncertainty	79.8 (15.1)	79.8 (22.0)	56.8 (16.2)	66.7 (11.8)	69.2 (19.2)	4.1	0.05	3.3	0.003	4.6	0.04
Isolation	76.8 (12.9)	87.5 (13.0)	66.5 (16.1)	68.8 (29.0)	74.2 (18.6)	2.0	0.17	2.4	0.03	3.4	0.08
Limitations	73.8 (27.4)	83.3 (12.7)	55.3 (23.7)	55.0 (19.2)	66.1 (24.2)	4.5	0.04	3.0	0.01	5.2	0.03
CDAT											
Overall	9.3 (1.7)	10.0 (1.5)	10.9 (2.2)	10.2 (2.9)	10.2 (2.1)	0.9	0.34	−1.4	0.17	1.2	0.28
Headaches and low energy	3.4 (1.0)	3.1 (1.1)	4.1 (0.9)	4.6 (2.7)	3.8 (1.4)	3.1	0.09	−1.9	0.06	0.4	0.54
Eaten gluten on purpose	1.1 (0.4)	1.3 (0.5)	1.0 (0.0)	1.0 (0.0)	1.1 (0.3)	1.7	0.20	2.0	0.05	2.9	0.10
	n (%)	n (%)	n (%)	n (%)	n (%)	χ^2	P	χ^2	P**		
Extremely vigilant	1 (14.3)	1 (14.3)	4 (36.4)	1 (20.0)	7 (23.3)	0.5	0.47	0.4	0.51		

*Higher CD-Specific Pediatric Quality Of Life (CDPQOL) sub-scores and overall scores suggest a higher degree of QOL; Scales 0–100.

[†]Higher Celiac Dietary Adherence Test (CDAT) scores suggest lower adherence.

[‡]Linear trend χ^2 and F, both d.f. = 1, χ^2 with continuity correction.

[§]Most adaptive + Adaptive versus Maladaptive + Most maladaptive, t-test d.f. = 28.

[¶]Most adaptive + Adaptive versus Maladaptive + Most maladaptive, controlling for age at enrollment, Main Effects F, d.f. = 1.

**Most adaptive + Adaptive versus Maladaptive + Most maladaptive, χ^2 with continuity correction, d.f. = 1.

to find acceptable gluten-free options when eating outside the home and felt confident to try new restaurants or food products that could accommodate their needs ('Gotten a lot easier over the years'). They used problem-focused coping strategies to reduce the risk of gluten contamination in a variety of settings. By contrast, adolescents with maladaptive eating behaviours were rigid in their GFD management, rarely willing to experiment with new restaurants or new food products ('I will only eat at trusted places'). Although these strategies ensured the avoidance of gluten, they did not remove the source of distress.

Trust versus avoidance
For adolescents with adaptive eating behaviours, the ability to trust others ensured inclusion in social events and maintenance of the GFD. These adolescents, although aware of gluten cross-contamination, managed this risk by asking questions and trusting the responses. They used external resources to help maintain their GFD ('I order GF and say it's for CD', 'Great Aunt makes sure I have something to eat – GF brownies, desserts – will call and ask if there is something that I want in particular'). By contrast, adolescents with maladaptive eating behaviours were heavily burdened by cross-contamination concerns when dining outside the home ('I worry about being out

and being contaminated') and anxiety and fear were prevalent in new situations. To manage, one adolescent said, 'I won't tell people I have coeliac disease . . . will offer to cook' preferring the safety of her own preparations; some would refuse to eat outside the home ('Don't go out to eat because of GF'). These adolescents controlled the risk of gluten contamination by using emotion-focused and internalising approaches, such as not attending school team/sporting functions involving food ('Ended up having to skip a summer program – there would be no GF assistance') and thoughts of family gatherings were angst filled ('I dread Thanksgiving').

Confidence versus controlling behaviour
Adolescents with adaptive eating behaviours had confidence in their ability to advocate for themselves and to maintain the GFD ('I use an index card with all the information'). If necessary, they would consume naturally gluten-free foods ('I bring my own fruit', 'chips, soda are fine'). They maintained a large social network of friends, classmates, family and others, who they made aware of their CD status, and who often advocated on their behalf ('My parents cook from scratch', 'Friends keep GF food for me'). By contrast, adolescents with maladaptive eating behaviours reported not informing their social network of

their CD diagnosis or gluten-free requirements ('Hard going out with friends when dining out'). They internalised their coping and did not use sources of support around them to maintain the GFD ('Don't eat others cooking', 'Stick to prepackaged food'). Instead, they felt a need to be in control of food situations and expected to inspect the kitchens where food would be prepared ('Have to go to restaurants on my downtime to take time to talk with the manager/chef'). They monitored food preparation and would visit or call restaurants in advance, thoroughly interview the staff on gluten cross-contamination measures ('A hawk – I notice everything in preparation'). For some, the perceived lack of control when eating outside the home often meant refusing events involving food and using avoidance strategies to manage their GFD ('Strictly GF home – no one allowed to bring anything gluten into home').

Awareness versus preoccupation
For adolescents with adaptive eating behaviours, maintaining a GFD was important but did not dominate daily thoughts outside of meal times ('Restaurant – not trustworthy – they'll be wrong about stuff and don't understand – but I'm still nice about it'). They reported becoming more aware of the GFD and how to manage it and reflected on the 'increasing amount of gluten-free products.' These adolescents were still aware of the risks of cross-contamination when eating outside the home ('Restaurants can make stupid mistakes') but accepted cross-contamination incidents and learned from them. By contrast, adolescents with maladaptive eating behaviours were preoccupied with future eating options ('I do a lot of research', 'You're always planning your meal'), spending hours researching locations or using Internet forums or mobile applications to determine whether a restaurant might be acceptable ('Look up restaurants ahead of time'). Some would make sure to eat before they went outside the home and, when cross-contamination did occur outside the home, were upset and dwelled on the incident ('I was upset that the [restaurant] staff not knowledgeable'). A summary of the various eating behaviour patterns and representative quotes is provided in the Supporting information, Table S1.

Discussion

Adolescents with CD develop different approaches to managing a GFD and different eating behaviours. Maladaptive eating behaviours associated with GFD management (i.e. those characterised by rigidity, avoidance, controlling behaviour and preoccupation) were reported by approximately half (53.3%) of our study sample. These maladaptive eating behaviours were described more often by older adolescents and were associated with diminished

QOL. Although maladaptive eating behaviour rates remain unknown, research suggests that adolescents with autoimmune disorders with a digestive component, such as CD, are at significantly increased risk of developing an eating disorder [31]. Estimates of adolescents with CD having or being affected by eating disorders are as high as 30% [19].

At present, it is unclear how GFD management approaches develop. Factors to consider in the development of maladaptive eating patterns include personality attributes, self-efficacy and ease of adapting to challenging situations, internality, coping strategies, CD and GFD knowledge of both self and others, and environmental and social support [11,35,42–44]. In addition, level of adherence to the GFD may also be a trigger [20]. Regardless of their specific antecedents, CD-related maladaptive eating behaviours deserve consideration. The relationships that we have described between specific eating behaviour patterns and CDPQOL can help inform the development of interventions to promote GFD adherence at the same time as preserving QOL.

All adolescents in the present study recognised the health benefits of maintaining a GFD. Several described symptoms (both gastrointestinal and nongastrointestinal) when exposed to gluten but, for most, these symptoms had not occurred recently. Although those in the maladaptive groups reported a significantly lower frequency of intentional gluten exposure, all groups had means closer to 1 (no exposure) than to 2 (one or two times in the past month) indicating good GFD management. Percentage of hypervigilance to gluten adherence, although more than double in the maladaptive group, was not significantly different. Again, we were constrained by a small sample size. The issue is worth further research because the trend is consistent with recent findings in another highly adherent adolescent population [45]. Although the total CDAT score did not differ across eating behaviour groups, the symptom-related items trended worse among the maladaptive groups. A measure of symptoms and perceived severity when gluten exposure occurs may have been more differentiating.

Adolescents with maladaptive eating behaviours described a more emotion-focused and internalising approach, expressed being nervous or anxious, spending a lot of time considering options, and relying on their own best judgment rather than involving others. These characteristics have striking similarities to personality and temperamental traits of those deemed to be at risk of diagnosable eating disorders (i.e. neuroticism, perfectionism and obsessiveness) [21]. In comparison, adolescents with more adaptive eating behaviours expressed a more problem-focused approach to maintaining the GFD. They relied on eating choices that tended to be naturally

gluten-free or used techniques, such as carrying instruction cards for restaurants to describe dietary restrictions. Their approach was more externalising, allowing others to assist and advocate for them.

Adolescents with maladaptive eating behaviours tended to be older [16.4 (1.0) years versus 14.7 (1.5) years] and older subjects tended to have lower CDPQOL scores. Older adolescents most likely have more occasion to navigate social situations involving eating outside of the home. With the onset of adulthood comes an increased responsibility for self-management [46].

We consider our findings that maladaptive eating behaviours were associated with lower QOL scores were not only statistically significant, but also clinically meaningful. For the adult version of the Celiac-Disease Quality of Life (CD-QOL) instrument [47], a difference of approximately 10 points on the CD-QOL scale was suggestive of clinical significance. A decline in 10 points on the CD-QOL scale was shown to be sufficient to move individuals into a worse category of self-rated health, psychological distress, functional status or pain [47]. For the adolescent version, there is no published data on the clinical significance of differences in the life CDPQOL scores. However, adolescents from the same population as the current study who participated in a later pilot study were asked (in addition to the CDPQOL items) to self-rate their QOL related to their CD on a scale of 1 = Poor, 2 = Fair, 3 = Good, 4 = Very Good or 5 = Excellent. Total CDPQOL and subscale scores were all linearly related across these self-rating categories. Most adolescents fell into the Excellent to Very Good range and average mean differences in CDPQOL scores between the Excellent and Very Good groups were generally about 15–20 points. In the present study, the differences in means between the two maladaptive eating behaviour groups were also on the order of 15–20 points. Thus, we feel confident that our findings suggest not just statistical significance, but clinical meaning as well.

Until research determines the exact 'strictness' of the GFD that will ensure healing of the mucosal lesion, CD patients will continue to be advised to maintain a strict GFD. The current guidance for periodic visits should be in accordance with current CD management recommendations [48,49]. Trained CD-specialist dietitians can assist with this counselling [48,49], with possible referrals to a psychotherapist to rule out eating concerns, including Avoidant/Restrictive Food Intake Disorder [21]. Encouraging family participation in care, including joining in person CD support groups, may help [48–50]. Consideration may be given to CD specific tools [51]. Parents, physicians and dietitians can all play a role in supporting positive eating behaviours and attitudes [12,18,19,46,48–50,52] (Table 3).

Limitations

This cross-sectional cohort study, investigating data from a larger study at an urban referral centre [36], presents results on a fairly homogenous sample. The findings may not be generalisable. Recall bias is inherent in the items that the participants choose to self-report. Participants did not necessarily offer responses that addressed all dimensions of our eating behaviour classification rubric; a structured interview may have allowed finer distinctions. Observational studies are another option for future research. The small sample size may have limited our ability to determine significance in certain results. Our symptom measure was based on just two items from the self-administered CDAT. A more extensive questionnaire on symptoms would have been preferable. Personality traits, parental psychosocial difficulties, including anxiety and depression, perceptions of costs and knowledge, as well as a diagnosable feeding or eating disorder, may also play a role in the relationship between eating behaviours and QOL, but were not

Table 3 Considerations for prevention of eating difficulties in adolescents with coeliac disease

Promote regular gastroenterologist visits and ongoing involvement of a registered dietitian nutritionist (RDN) specialised in coeliac disease (CD), beyond the initial diagnosis [48,49]
Through conversations over time, practitioners should promote increased gluten-free diet (GFD) adherence/reduce inadvertent gluten exposures, correct any nutritional deficits, promote normal growth and development, and monitor for continued or new symptoms, including other digestive-related morbidities [48,49]
Upon diagnosis, patients should be encouraged to join a CD support groups [48–50] particularly in-person if possible [50]
Encourage family members involvement as part of the ongoing support team [48,49]
RDNs should assess whether a patient's approach to GFD adherence may interfere with daily activities and quality of life
CD specific tools may be useful in assessments. The newly developed the coeliac disease food attitudes and beliefs scale, validated in adults, may help in assessing CD-Specific food-related behaviours, although its utility with adolescents and for clinical practice is unknown [51]
A nonpunitive approach to dietary counselling should be adopted [24,25]
If the treatment team has concerns about a patient's eating approach that is not resolving, the patient should be referred to an eating disorder specialist (psychiatrist, psychologist, social worker) for further evaluation
Psychiatric co-morbidities such as anxiety or depression should be identified, and appropriate referral undertaken

measured in this study [21,52]. Despite these limitations, to our knowledge, this is the first mixed methods analysis that describes the approach towards maintaining a GFD, highlighting the need for additional eating behaviour research among adolescents with CD.

Conclusions

Approximately half (53.3%) of the adolescents with CD in this study, expressed maladaptive eating behaviours in managing a GFD. Maladaptive eating behaviours were characterised by rigidity, avoidance, controlling behaviour and preoccupation, all known risk factors for the development of diagnosable eating disorders. Maladaptive behaviours in this study sample were associated with diminished CDPQOL. Practitioners should encourage patients and their families to pursue ongoing follow-up with themselves as well as a trained CD-specialised dietitian to promote GFD adherence to a GFD in a way that promotes higher QOL with positive behavioural approach to GFD management, providing psychosocial support, as warranted. Early intervention may prevent further complications. Our results highlight the vulnerability of adolescent CD patients as they transition into adulthood [46] when they may seek less medical care for their CD. It is during this transition period, when adolescents are susceptible to the aggravation of maladaptive eating patterns, that support may be most needed.

Transparency declaration

The lead author affirms that this manuscript is an honest, accurate and transparent account of the study being reported. The reporting of this work is compliant with STROBE [53] guidelines. The lead author affirms that no important aspects of the study have been omitted and that any discrepancies from the study as planned have been explained.

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Supporting information

Additional supporting information may be found online in the Supporting Information section at the end of the article.

Appendix S1. Coding matrix.

Table S1. Lived experience patterns and quotes by eating behaviour.