Original Article

Self-Report of Adherence to Gluten-Free Diet in Patients with Celiac Disease Versus Expert Evaluation

Ali Jafari[1,2], Fazel IsapanahAmlashi[1,2], Zahra Norouzi[1,2], Iman ShahabiNasab[1,2*], Sima Besharat[2,3], Puria Qadirian[1,2]

[1]Student Research Committee, Department of Nutrition, School of Health, Golestan University of Medical Sciences, Gorgan, Iran
[2]Golestan Research Center of Gastroenterology and Hepatology, Golestan University of Medical Sciences, Gorgan, Iran
[3]Clinical Research Development Unit (CRDU), Sayyad-e-Shirazi Hospital, Golestan University of Medical Sciences, Gorgan, Iran

ABSTRACT

Background
Consuming gluten can lead to the immune-mediated condition known as celiac disease (CD) in genetically-prone people. Presently, the only approved and available treatment is stringent and lifetime devotion to a gluten-free diet (GFD). This study aimed to assess GFD adherence in patients with CD in Golestan province, northeast Iran.

Materials and Methods
All cases with confirmed CD registered in the Golestan Registry of Celiac (N=220) were selected for this cross-sectional study. 87 patients volunteered to take part in the interviews and completed the questionnaire. Since the day of diagnosis, all patients in this center had been treated with a GFD, but their adherence to the regime was unknown. Celiac Dietary Adherence Test (CDAT) and the Standardized Dietician Evaluation (SDE) were used to evaluate their adherence to GFD.

Results
Among the 85 individuals who responded in depth to the CDAT, the mean (standard deviation) age was 32.41 (15.45) years, 32 (36.8%) were male, and 72 (32.8%) adhered to the diet according to their self-expression. However, in the SDE, only 52 (30.23%) exhibited great to good adherence.

Conclusions
The adherence and weak adherence groups had similar mean ages. However, non-adherence was associated with higher mean age. Although women had stronger adherence, there was no difference in sex. The elderly and males exhibited low GFD adherence.

Keywords: Gluten-free diet, Celiac disease, Adherence

Please cite this paper as:
Jafari A, Isapanah Amlashi F, Norouzi Z, Shahabi Nasab I, Besharat S, Qadirian P. Self-report of adherence to gluten-free diet in patients with celiac disease versus expert evaluation. *Govaresh* 2023;28: 126-130.

Corresponding author:
Iman ShahabiNasab, MD
Golestan Research Center of Gastroenterology and Hepatology,
3rd floor, Heart Complex, Sayyad-e-Shirazi Hospital,
Sayyad-e-Shirazi Boulevard, Gorgan city, Golestan province, Iran.
Telefax: +981732251910
Email: shahabnasabi@gmail.com

Received: 09 Jan. 2023
Revised: 11 May 2023
Accepted: 12 May 2023

INTRODUCTION

Celiac disease (CD), an immune-mediated disorder caused by the digestion of gluten-containing foods in genetically predisposed individuals, occurs in almost 1% (0.3-1.3%) of the world's population.

It is estimated to report 1% to 3% of people screened for CD through serology or biopsy (1). CD often leads to impaired nutrient absorption due to the remarkable atrophy and loss of the small intestinal villi. Complete elimination of gluten from the diet is strongly suggested to resolve symptoms, heal the villi and prevent the consequences (1,2).

Adherence to a gluten-free diet (GFD) leads to the regeneration of intestinal villi after 6 to 24 months. GFD ensures longevity and relief from abdominal pain, bloating, loose stools, constipation, iron deficiency anemia, short stature, and osteoporosis. It also reduces the risk of cardiovascular disease and intestinal tumors in adults with CD (1,3).

GFD means eliminating wheat, rye, and barley products and processed cereal products. These may be replaced by natural, gluten-free products (corn, rice, oats, buckwheat, meat, fish, vegetables, and fruits) or products from which gluten has been eliminated (3).

But unfortunately, 20-50% of patients have no dietary restrictions (4,5). Most patients on GFD do very well; despite the diet being troublesome in terms of cost (6), nutritional value (7), and social constraints (8-10).

Previous studies reported that even those individuals who strongly believe that they are following a GFD, do not completely identify foods with gluten and so continue to consume gluten (11).

In a recent study by Głady et al., as many as 24-52% of patients with CD did not adhere well enough to GFD, which shows a significant problem in these patients (12).

Various factors may affect adherence, especially age at diagnosis (13). It means that a longer duration of the disease leads to poorer adherence. In a study by Pedoto et al. on children with CD, a decrease in adherence to GFD was seen during the follow-up (14).It has also been reported that self-reported adherence (by patients or parents) overestimates the scores and misleads the evaluations, and there is an essential need for regular assessment and education by an expert dietician (15).

This study aimed to evaluate adherence to GFD in patients with CD from the patients' point of view in comparison to the evaluation performed by the specialist.

MATERIALS AND METHODS

Study population

In our study, all CD cases recorded in the Golestan Registry of Celiac (N=220) were recruited from the Golestan Research Center of Gastroenterology database Hepatology (GRCGH). A biopsy of the small intestine confirmed the diagnosis of CD. Marsh is graded from one to three, which includes Marsh 1 with almost normal mucus except intraepithelial lymphocyte infiltration, Marsh 2 with the additional presence of crypt hypertrophy, and Marsh 3 with flattening of the mucosa due to the so-called villous atrophy and swelling of the lamina propria.

87 patients agreed to participate in the interviews and filled out the questionnaire. All patients in this center were under treatment with a GFD from the day of diagnosis, but their adherence to the regime was unclear.

After explaining the research project and obtaining written consent from patients, questionnaires were completed. The questionnaires included demographic information (age, sex, educational level, duration of the disease, method of diagnosis, and family history).

Adherence to GFD was evaluated byCeliac Disease Adherence Test (CDAT), and Standardized Dietician Evaluation (SDE).

CDAT has seven meaningful and straightforward questions, and scores are summed to reach the final score (ranges 7-35). According to the classification of Nikniaz et al., the final scores were divided into three groups: good adherence (less than 13), moderate adherence (13 to 17), and poor adherence (more than 17). The reliability and validity of the Persian version of CDAT were confirmed in the previous study by Nikniaz et al.(16). In this study, we considered reasonable and moderate as "adherence" and poor as "non-adherence" in the final analysis.

Also, the Standardized Dietician Evaluation (SDE) was used to assess the patient's adherence from the interviewer's perspective. Trained colleagues interviewed patients to complete the questionnaires. According to the questionnaire of Leffler et al., during the interview, several questions were answered from the questioner's point of view. Patients were classified through the Likert scoring criteria from 1 (perfect GFD adherence) to 6 (no GFD adherence) (17). In the final analysis, we grouped fair, poor, and very poor as "non-adherence" and perfect and good as "adherence".

Patients were divided into two categories: under 19 years old as adolescents and over 19 as adults.

Besides, they were asked if resolving symptoms occurred after consuming gluten-containing substances and controlling the anti-tTg titer at least four times a year.

Statistical analysis

After gathering data from both questionnaires, the assumption of normality was verified with the Kolmogorov–Smirnov test. Results of the categorical variables were shown as percentages, mean and standard deviation, or median and interquartile range, where appropriate. A $P<0.05$ was considered to be statistically significant.

Comparisons between the two groups were assessed with a Mann–Whitney test or Chi-square tests, as appropriate. Statistical processing was performed using the statistical software SPSS v 16.0.

Ethical consideration

The local EthicsCommittee of Golestan University of Medical Sciences approved the study protocol (IR.GOUMS.REC.1398.310). The purpose of the study was explained to all participants, and informed consent was taken from all.

RESULTS

In this cross-sectional study, 87 patients with CD, a mean (SD) age of 32.41 (15.45) years, and 63.2% female were interviewed using the questionnaires as mentioned earlier. Among them, 85 completed the CDAT questionnaire, and 86 answered the questions to calculate their SDE score (Table 1).

According to Table 2 (re-classification of the CDAT score), 15.3% claimed they did not adhere to the GFD tightly. However, when interviews were followed by more detailed questions by a trained nutritionist or general practitioner (SDE score), 39.5% showed non-adherence.

Analysis showed no statistically significant differences between the CDAT and SDE score and demographic variables such as sex, age, age at diagnosis, and educational degree ($P>0.05$).

According to SDE,there was no significant difference in mean age between the two groups with adherence (30.23 ± 16.16 years) and non-adherence (35.53 ± 14.11 years). The mean age was generally higher in the non-adherence group, though. Regarding sex, there was no significant difference between the two groups, even though women generally had higher levels of adherence (F/M was 35/17 in adherence group and 20/14 in non-adherence).

Table 1. Basic characteristics of the study population with CD

Variable		Measure
Age, mean (SD), years		32.41 (15.45)
Age at diagnosis, mean (SD), years		24.66(15.15)
Sex, N (%)	Male	32 (36.8)
	Female	55 (63.2)
Age group, N (%), years	<=19	23 (26.4)
	>19	64 (73.6)
Marsh classification, N (%)	<3	32 (36.7)
	≥3	55 (63.2)

Table 2. Scores classification from CDAT and SDE questionnaires considering the adherence to GFDin patients with CD

Variable	classifications	Number (%)
CDAT* score	Good	47 (55.3)
	Moderate	25 (29.4)
	Poor	13 (15.3)
SDE† score	perfect	35 (40.7)
	good	17 (19.8)
	fair	22 (25.6)
	poor	5 (5.8)
	very poor	7 (8.1)

*CDAT=Celiac Disease Adherence Test; † SDE=Standardized Dietician Evaluation

DISCUSSION

Results of the present study showed differences between self-reported gluten-free adherence and expert view. Although the primary treatment strategy for CD is tight adherence to GFD, some adults in this study had sub-optimal diet compliance, as shown in previous studies (12,18-20).

Determination of adherence to GFD is methodologically challenging (21), so combining CDAT scores and SDE based on the interviewer's perspectives is known to evaluate it better than a stand-alone test (22). Our results showed that 60.5% (in SDE) and 84.7% (in CDAT) of patients adhered enough to GFD.

As previously shown in other studies, most celiac patients overestimate their adherence (11,12), and there are discrepancies between what they name a strict gluten-free regimen and what actually happens.

Some patients are unaware of the gluten hidden in

various sources, such as processed meats, makeup & body care, medication & supplements, sauces, etc. (23). This can be one of the reasons for the difference between CDAT and SDE results.

Our results showed that those with better adherence are younger and in the very early years of their diagnosis. It means that when someone is younger, and CD is newly diagnosed, there are more cautious about foods. This issue was reported in other previous studies as well (14).

One of the main reasons our patients claimed for not adhering to GFD was the high cost and difficulty in finding gluten-free foods and products, the same as in other parts of the world (9,24). In addition, they complained about other factors such as the unpleasant taste of gluten-free substances, troublesome diet, inability to read food labels, inability to eat outdoors, ridicule of others, and lack of social sympathy.

In the study by Halmos et al. (24), those who were less able to read labels were more likely not to identify gluten-free foods correctly, but there was an over-restriction of diet in this group that resulted from poor knowledge.

In our study, the participants' low level of education and knowledge resulted in the difference between adherence from the expert's point of view and self-reports. It means that patients with CD think that they have good or excellent adherence and avoid gluten so well. However, when an expert mentions the other possible ways of taking gluten and categorizes the adherence more strictly, it reveals that many more important points must be considered.

It should be suggested to clinicians to pay more attention to thorough consultations and active interactions with patients with CD and to health policymakers to improve the general awareness about the content of gluten in foods.

This study has its limitations. The study's small sample size made it difficult to determine the exact adherence rates and predictors. Several patients could not attend the GRCGH themselves, and one of their relatives answered the questionnaires on their behalf, which may affect the responses and disrupt the interpretations.

CONCLUSION

This study revealed that our patients with CD had generally poor GFD adherence, particularly the elderly and males.

ACKNOWLEDGMENTS

Authors tend to thank the Deputy of Research, Golestan University of Medical Sciences, for its financial support, and the personnel of GRCGH for their collaborations.

AUTHORS CONTRIBUTION

I.SH, F.I.A, and P.Q: Substantial contributions to the conception, drafting the work or revising it critically for important intellectual content, final approval of the version to be published, agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

S.B: Substantial contributions to the design of the work, drafting the work or revising it critically for important intellectual content, final approval of the version to be published, agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

A.J and Z.N: Substantial contributions to the interpretation of data for the work, drafting the work or revising it critically for important intellectual content, final approval of the version to be published, agreement to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved. All authors have read and agreed to the published version of the manuscript.

COMPETING INTERESTS

The authors declare no conflict of interest related to this work.

FUNDING

This research was funded by Golestan University of Medical Sciences, Deputy of Research.

REFERENCES

1. Husby S, Koletzko S, Korponay-Szabó IR, Mearin ML, Phillips A, Shamir R, et al. European Society for Pediatric Gastroenterology, Hepatology, and Nutrition guidelines for the diagnosis of coeliac disease. *J Pediatr Gastroenterol Nutr* 2012;54(1):136-60. doi: 10.1097/MPG.0b013e31821a23d0
2. Silvester JA, Rashid M. Long-term management of patients with celiac disease: current practices of gastroenterologists in Canada. *Can J Gastroenterol* 2010;24(8):499-509. doi: 10.1155/2010/140289
3. Bathrellou E, Kontogianni MD, Panagiotakos DB. Celiac

disease and non-celiac gluten or wheat sensitivity and health in later life: a review. *Maturitas* 2018;112:29-33. doi: 10.1016/j.maturitas.2018.03.014
4. Ciacci C, Cirillo M, Cavallaro R, Mazzacca G. Long-term follow-up of celiac adults on gluten-free diet: prevalence and correlates of intestinal damage. *Digestion* 2002;66(3):178-85. doi: 10.1159/000066757
5. Högberg L, Grodzinsky E, Stenhammar L. Better dietary compliance in patients with coeliac disease diagnosed in early childhood. *Scand J Gastroenterol* 2003;38(7):751-4. doi: 10.1080/00365520310003318
6. Lee AR, Ng DL, Zivin J, Green PH. Economic burden of a gluten-free diet. *J Hum Nutr Diet* 2007;20(5):423-30. doi: 10.1111/j.1365-277X.2007.00763.x
7. Thompson T, Dennis M, Higgins LA, Lee AR, Sharrett MK. Gluten-free diet survey: are Americans with coeliac disease consuming recommended amounts of fibre, iron, calcium and grain foods? *J Hum Nutr Diet* 2005;18(3):163-9. doi: 10.1111/j.1365-277X.2005.00607.x
8. Addolorato G, De Lorenzi G, Abenavoli L, Leggio L, Capristo E, Gasbarrini G. Psychological support counselling improves gluten-free diet compliance in coeliac patients with affective disorders. *Aliment Pharmacol Ther* 2004;20(7):777-82. doi: 10.1111/j.1365-2036.2004.02193.x
9. Leffler DA, Edwards-George J, Dennis M, Schuppan D, Cook F, Franko DL, et al. Factors that influence adherence to a gluten-free diet in adults with celiac disease. *Dig Dis Sci* 2008;53(6):1573-81. doi: 10.1007/s10620-007-0055-3
10. Sverker A, Hensing G, Hallert C. 'Controlled by food'- lived experiences of coeliac disease. *J Hum Nutr Diet* 2005;18(3):171-80. doi: 10.1111/j.1365-277X.2005.00591.x
11. Silvester JA, Weiten D, Graff LA, Walker JR, Duerksen DR. Is it gluten-free? Relationship between self-reported gluten-free diet adherence and knowledge of gluten content of foods. *Nutrition* 2016;32(7-8):777-83. doi: 10.1016/j.nut.2016.01.021
12. Gładyś K, Dardzińska J, Guzek M, Adrych K, Małgorzewicz S. Celiac dietary adherence test and standardized dietician evaluation in assessment of adherence to a gluten-free diet in patients with celiac disease. *Nutrients* 2020;12(8):2300. doi: 10.3390/nu12082300
13. Freeman HJ. Detection of adult celiac disease with duodenal screening biopsies over a 30-year period. *Can J Gastroenterol* 2013;27(7):405-8. doi: 10.1155/2013/347902
14. Pedoto D, Troncone R, Massitti M, Greco L, Auricchio R. Adherence to gluten-free diet in coeliac paediatric patients assessed through a questionnaire positively influences growth and quality of life. *Nutrients* 2020;12(12):3802. doi: 10.3390/nu12123802
15. Dowhaniuk JK, Mileski H, Saab J, Tutelman P, Thabane L, Brill H. The gluten free diet: assessing adherence in a pediatric celiac disease population. *J Can Assoc Gastroenterol* 2020;3(2):67-73. doi: 10.1093/jcag/gwy067
16. Nikniaz Z, Asghari Jafarabadi M, Ghaffarifar S, Saeedi Z, Akbari Namvar Z, Shirmohammadi M. Psychometric properties of the Persian version of the celiac disease adherence test questionnaire. *BMC Gastroenterol* 2020;20(1):247. doi: 10.1186/s12876-020-01396-8
17. Leffler DA, Dennis M, Edwards George JB, Jamma S, Magge S, Cook EF, et al. A simple validated gluten-free diet adherence survey for adults with celiac disease. *Clin Gastroenterol Hepatol* 2009;7(5): 530-6.e2. doi: 10.1016/j.cgh.2008.12.032
18. Cadenhead JW, Wolf RL, Lebwohl B, Lee AR, Zybert P, Reilly NR, et al. 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. *J Hum Nutr Diet* 2019;32(3):311-20. doi: 10.1111/jhn.12638
19. Johansson K, Norström F, Nordyke K, Myleus A. Celiac dietary adherence test simplifies determining adherence to a gluten-free diet in Swedish adolescents. *J Pediatr Gastroenterol Nutr* 2019;69(5):575-80. doi: 10.1097/mpg.0000000000002451
20. Mehta P, Pan Z, Riley MD, Liu E. Adherence to a gluten-free diet: assessment by dietician interview and serology. *J Pediatr Gastroenterol Nutr* 2018;66(3):e67-e70. doi: 10.1097/mpg.0000000000001705
21. Ludvigsson JF, Bai JC, Biagi F, Card TR, Ciacci C, Ciclitira PJ, et al. Diagnosis and management of adult coeliac disease: guidelines from the British Society of Gastroenterology. *Gut* 2014;63(8):1210-28. doi: 10.1136/gutjnl-2013-306578
22. Hlywiak KH. Hidden sources of gluten. *Pract Gastroenterol* 2008;32:27-39.
23. Villafuerte-Galvez J, Vanga RR, Dennis M, Hansen J, Leffler DA, Kelly CP, et al. Factors governing long-term adherence to a gluten-free diet in adult patients with coeliac disease. *Aliment Pharmacol Ther* 2015;42(6):753-60. doi: 10.1111/apt.13319
24. Halmos EP, Deng M, Knowles SR, Sainsbury K, Mullan B, Tye-Din JA. Food knowledge and psychological state predict adherence to a gluten-free diet in a survey of 5310 Australians and New Zealanders with coeliac disease. *Aliment Pharmacol Ther* 2018;48(1):78-86. doi: 10.1111/apt.14791