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Alimentary Tract

Sequence of acquisition of self-management skills to follow a gluten-free diet by adults with celiac disease

Emma M. Clerx[a,b], Jocelyn Silvester[a,c,d,1], Daniel Leffler[a], Maya DeGroote[c], Laurie N. Fishman[d,*]

[a] Celiac Center, Beth Israel Deaconess Medical Center, United States
[b] Harvard University, United States
[c] Celiac Center, Boston Children's Hospital,United States
[d] Division of Gastroenterology, Boston Children's Hospital, United States

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ABSTRACT

Background: Celiac disease (CD) treatment is lifelong adherence to a gluten-free diet (GFD), requiring mastery of numerous skills to maintain health.
Aims: To assess the rate of self-management skill acquisition following diagnosis, and the influence of demographic factors on this rate.
Methods: Patients attending a celiac center were invited to complete an anonymous survey which reported demographic information and time for mastery of self-management skills relevant to CD.
Results: Completed surveys were returned by 137 patients (79% female). Most participants reported mastering skills that involved identifying gluten-containing versus gluten-free foods within 6 months. Explaining CD and GFD to others required 1–2 years. Identifying gluten in medications and supplements required 3–5 years. Traveling internationally with GFD adherence required more than five years to learn. Demographic factors were not associated with the rate of acquisition.
Conclusions: This is the first description of a timeline for relevant skill acquisition following diagnosis for CD. A sequence emerges, with most patients learning skills relevant to home, then social settings, then the workplace, and, finally, unfamiliar settings. Awareness of this progression of mastery, and particular recognition of difficult skills will allow physicians and dietitians to provide CD patients with targeted education and resources to facilitate adherence.

1. Introduction

Celiac disease (CD) is a chronic immune-mediated enteropathy perpetuated by the ingestion of gluten [1]. The world-wide prevalence of CD is approximately1% [1] with regional variation [2,3], and the only treatment is lifelong adherence to a gluten-free diet (GFD) [4–6]. Patients whose treatment is focused on dietary intervention, such as in the case of CD, food allergy, and diabetes, are faced with a complex set of skills to master following diagnosis in order to manage their disease adequately. Patients with CD must learn a variety of self-management skills in order to safely maintain a GFD and avoid cross-contamination in restaurants and during domestic and international travel. Other skills that CD patients must learn include identifying potential sources of gluten in supplements and medications. Adherence can be quite challenging for CD patients because many do not experience symptoms immediately after gluten ingestion [7].

It is important that CD patients master these self-management skills because persistent gluten exposure is associated with further health complications, including osteopenia [8], lymphoma [9], and increased mortality rate [10,11]. Despite the marked improvements in health and quality of life linked to GFD adherence, many patients fail to adhere fully to the GFD. While some gluten exposure may be intentional, in other cases gluten exposure may be "accidental" due to either knowledge gaps or lack of planning [12].

The timeline of learning the self-management skills necessary for GFD adherence following diagnosis of CD is unknown. Thus, the primary aim of this study was to gather the necessary information from patients in order to outline the progression of learning following diagnosis and the recommendation to follow a GFD. A secondary aim was to identify demographic factors that may influ-

* Corresponding author.
E-mail address: laurie.fishman@childrens.harvard.edu (L.N. Fishman).
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ence the rate of learning. Knowledge of the rate of mastery of self-management skills in CD patients, and any relevant demographic factors, would aid physicians and dietitians in treating CD patients. This information can provide insight into areas where targeted patient education is needed in order to facilitate CD management and GFD adherence.

2. Methods

2.1. Survey design

An expert panel consisting of gastroenterologists, dietitians, and individuals with CD was assembled to discuss the various self-management skills necessary to maintain health with CD, as well as the behavioral factors influencing GFD adherence. Over a series of meetings, a self-administered survey was created that included items related to the time required to master various skills relating to GFD adherence using a categorical scale (<6 months, 6 months to <1 year, 1–2 years, 3–5 years, >5 years, Not Yet, Do not Know, or Choose Not To). The survey underwent several rounds of pilot testing with celiac patients, some of who were also clinicians, and revisions clarified ambiguities raised. The final instrument contained 17 items, including: identification of foods that may contain gluten, explaining CD and a GFD to others, ability to eat safely at home and in social settings outside the home, ability to identify medications and supplements as GF, and ability to assess risk of gluten exposure and take necessary precautions to maintain health and GFD adherence while traveling. The survey also included items related to symptoms of gluten exposure and the diagnostic process.

Participants were asked to report if any family or household members had CD, diabetes, or food allergies, as this might heighten awareness of diet prior to the diagnosis and accelerate the learning process. Willing participants also provided relevant demographic information, including marital status, education level, employment status, and occupation in order to assess for any correlations between demographic factors and duration of time needed to master self-management skills. Patients were invited to provide any additional written comments upon completion of the survey. This study was approved by the Beth Israel Deaconess Medical Center (BIDMC) Committee of Clinical Investigations (CCI).

2.2. Recruitment

Patients attending the outpatient clinic at the Celiac Center at BIDMC between July 2016 and June 2017 were invited to complete the survey. Exclusion criteria were: age less than 18 years, inability to complete a written survey in English, significant cognitive impairment, or lack of CD diagnosis. Participants completed the survey in clinic. A letter introducing the survey explained that consent was implied by submission of a completed survey to allow respondents to remain anonymous.

2.3. Analysis

Data analysis was performed using R [13] in R studio [14]. Survey responses that characterized the study population were analyzed using descriptive statistics. Participants' occupation types were coded and categorized according to the Bureau of Labor Statistics' major classification principles [15] with the addition of categories: student, retired, and housewife/stay-at-home mom to better characterize participants, so that further analysis could be conducted. Wilcoxon signed-rank tests were performed to assess for differences in rates of learning various skills based on gender, marital status, and varying family history patterns.

Table 1
Characteristics of the study population.

	Patient cohort studied
Median current age (IQR) (n = 137)	38 (28–58)
Median age at diagnosis (IQR) (n = 137)	35 (23–50)
Female % (n)	79% (107)
Median age at first celiac symptoms (IQR)	23 (23–43)
Median years on GFD (IQR)	5 (2–7)
Diagnosis Type: (n = 135)	
% Diagnosed with only intestinal biopsy (n)	17% (23)
% Diagnosed with only blood antibody tests (n)	13% (18)
% Diagnosed using both (n)	70% (94)
Family history of related autoimmune disorder: (n = 77)	
% With household member with celiac disease (n)	23% (31)
% With household member with diabetes (n)	15% (21)
% With household member food allergies (n)	19% (25)
Marital status: (n = 131)	
% Single (n)	42% (55)
% Married (n)	51% (67)
% Divorced (n)	6% (8)
% Widowed (n)	1% (1)
Education level: (n = 134)	
% Less than high school (n)	1% (2)
% High school or trade school graduate (n)	3% (4)
% Some college, associate, or bachelor's degree (n)	63% (84)
% Graduate or professional degree (n)	33% (44)
Employment status: (n = 126)	
% Full-time (n)	60% (76)
% Part-time (n)	18% (22)
% Retired (n)	7% (9)
% Unemployed (n)	15% (19)
Occupation Type[a]: (n = 113)	
% Management, professional, and \\\Related Occupations (n)	64% (72)
% Service occupations (n)	4% (4)
% Sales and office occupations (n)	8% (9)
% Production, transportation, and///material moving occupations (n)	2% (2)
% Stay-at-home-mom or housewife///(n)	2% (2)
% Student (n)	13% (15)
% Retired (n)	8% (9)

[a] Occupation type was determined according to the Bureau of Labor Statistics' Classification Principles with the addition of Stay-at-Home Mom or Housewife, Student, and Retired.

3. Results

Completed surveys were returned by 137 patients with CD with a median age of 38 years (IQR 28–58; Table 1). The majority of participants reported biopsy-proven CD (87%) diagnosed at a median age of 35 years (IQR 23–50), and the median years that participants had spent on the GFD was 5 (IQR 2–7). Participants were primarily fully-employed (60%), married (51%), and female (79%). Many participants reported a household member with a related disease in which dietary modification is a key component of self-management, including celiac disease (23%), diabetes (15%), and food allergies (19%).

The majority of participants (82%) experienced gastrointestinal symptoms in response to gluten exposure, including altered bowel habits, weight loss, abdominal pain, bloating, nausea or vomiting, and flatulence (Table 2). Nearly 50% of participants experienced non-classical symptoms, including fatigue, depression, anxiety, and mood swings.

The survey responses showed an array of skills requiring varying amounts of time for patients to master (Fig. 1). Skills involving the assessment of basic food safety, such as identifying unsafe, gluten-containing foods and recognizing the GF symbol as 'Gluten-Free', were associated with the least amount of time with the majority of participants reporting that these tasks were mastered within 6 months. Cleaning off shared eating spaces, washing hands before meals, and recognizing and describing the effects of gluten on

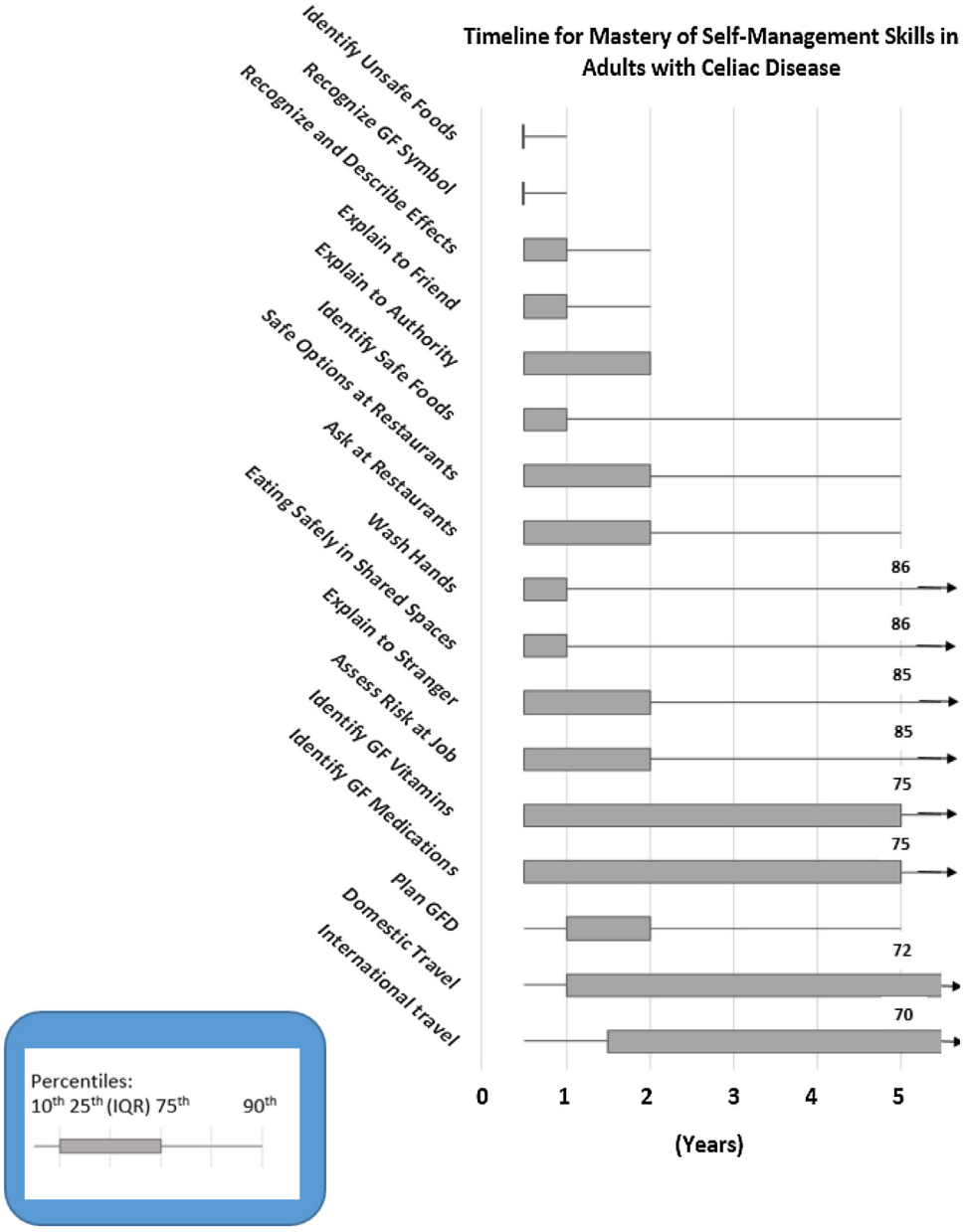

Fig. 1. This figure visually demonstrates the time in years of mastery of self-management skills by adults with celiac disease. The grey bar demonstrates the 25–95 percentiles for each skill.

the body required longer amounts of time, with most participants reporting that these tasks required six months to one year to learn. Most participants noted that learning how to explain CD and the GFD to others with no prior knowledge of CD, asking about gluten-free food at restaurants, and assessing risk of gluten exposure outside the home required one to two years to master. Identifying medications, vitamins, or supplements as gluten-free required three to five years for most participants to learn. Tasks involving assessing the risk of gluten exposure during domestic and international travel required the longest amounts of time to master—with most participants reporting that these skills required more than five years to fully learn, or they had not yet mastered them.

Demographic factors did not seem to have any influence on rate of skill acquisition. Wilcoxon signed-rank tests assessing for differences in rates of learning various skills based on gender, marital status, and varying family history patterns yielded no significant findings, with all p-values greater than 0.05. Specifically, there were no significant differences in learning rates between male versus female participants; married versus single versus divorced participants; nor participants with a family history of celiac disease versus diabetes versus food allergies versus no relevant family history pattern. Education and occupation were not diverse enough to allow for comparison.

4. Discussion

This observational study provides valuable information regarding a variable timeline for CD patients' acquisition of skills required for successful adherence to the GFD and proper disease management. It does appear that CD patients develop the skills to manage their health and adherence to the GFD in a clear progression: first mastering skills in the home, then in social settings, then in the workplace, and finally in unfamiliar settings, such as those encountered during travel. Knowledge of this characteristic developmental progression will allow physicians, dietitians, and other health professionals to assess CD patients' level of comfort with the GFD and

Table 2
Respondents symptoms.

Symptoms to gluten exposure (n = 137 participants)	% who experienced each symptom
Altered bowel habits (n)	61% (84)
Abdominal pain (n)	28% (38)
Fatigue (n)	24% (33)
Bloating (n)	22% (30)
Nausea or vomiting (n)	13% (18)
Weight loss (n)	12% (16)
Flatulence (n)	12% (16)
Depression, anxiety, or mood swings (n)	12% (16)
Anemia (n)	11% (15)
Headaches/migraines (n)	9% (12)
Brain Fog (n)	7% (9)
Joint pain (n)	6% (8)
Dermatitis herpetiformis (DH) (n)	5% (7)
Bone disease (n)	4% (5)
"None" (n)	3% (4)

Percentage of respondents who experience each symptom when accidentally or intentionally ingesting gluten.

note certain milestones in mastery of their treatment in order to facilitate disease management. However, a long mastery time may reflect less on the skills of the patient and more on the lack of appropriate education by the providers. For example, not all patients are taught about searching for gluten in prescription medication, but may learn through extensive peer networks.

This progression must be taken as an optimistic view of GFD learning. It is known that patients often overestimate their adherence to the GFD when compared to assessment by a trained CD dietitian [16]. Adherence is difficult, as is evidenced by the 20–50% of CD patients who report inadequate GFD compliance [17,18]. The prolonged time required to acquire self-perceived competence in the skills required to follow a GFD has significant clinical implications. Studies of so-called "non-responsive" CD often consider patients with ongoing symptoms after 6–12 months on a GFD to be "non-responsive" [19]. The results of the current study suggest that this diagnosis may be premature as many of these patients have not yet acquired the necessary skills to consistently follow a GFD. Thus, it is not surprising that the most common cause of "non-responsive" celiac disease is gluten exposure [20]. Similarly, a further considered assessment of adherence that incorporates the timeline of skill acquisition may also guide the appropriate timing of repeat biopsies.

Our study also has implications for patient education. Patients and even providers may expect that a single dietary educational session regarding gluten-containing foods would allow patients to follow a GFD. Our findings show that patients master some tasks quickly, but that there is a hierarchy of tasks that may require returning for further education or support, particularly related to following a GFD outside the relatively controlled home setting. It also allows providers to take a more sophisticated view of adherence, as patients lacking some of the more complex skills may follow a GFD at a great personal or social cost. Many CD patients perceive the burden of their disease treatment to be significant, comparable to that of end stage renal disease [21] and report a poor overall quality of life [22]. This may be a manifestation of the increased cost [23] and social constraints [24] associated with following a GFD. In surveys of patients with CD, nearly all report avoiding travelling or going to restaurants because of a GFD [25]. Development of the more advanced self-management skills needed to follow a GFD while traveling or socializing outside the home may help improve the perceived quality of life.

The average patient with celiac disease may get far less education than the respondents. Our patients receive extensive information and counseling related to the gluten-free diet, reading labels, recognizing potential sources of gluten exposure, other health problems potentially associated with celiac disease, a variety of resources addressing celiac-specific concerns, including an introduction to a local patient support group. We recommend three dietitian visits during the first year after diagnosis and content is tailored to patient needs. The initial visit tends to focus on gluten, ingredients and label-reading and cross contamination, whereas subsequent visits tend to focus on areas of concern or challenge for the patient, typically cross-contamination and eating out. The majority of these visits are with the celiac center dietitian.

Limitations of this study include its observational and retrospective nature which makes it subject to selection and recall bias. Self-assessment of skill acquisition may have been confounded by varying degrees of self-confidence and differing personality types of the respondents. The mean age of the study cohort was younger than the general clinic and thus might skew the results. The study population of highly-educated managers and professionals followed at a tertiary care facility would be more likely to underestimate time to skill acquisition as these individuals might spend more effort and time learning health related skills. On the other hand, they may hold themselves to very high internal standards. Further studies, with greater socioeconomic diversity, can explore how different populations view and achieve health task mastery.

The timeline for acquisition of self-management skills that CD patients must master to successfully follow a GFD, presented in this paper, provides a useful tool to monitor the progress in treatment of patients with CD. Physicians' and dietitians' knowledge of these milestones will benefit patients and help to facilitate their disease management. Targeted education has proven effective in other diseases, such as diabetes and food allergy, for which management entails dietary vigilance [26]. There is also evidence that CD patients who are more knowledgeable about CD and the GFD adhere better to the diet [27–29]. Therefore, improved education and resources for CD patients, especially in areas that have been highlighted as difficult to master, are needed to empower CD patients as they learn the skills required to follow a GFD. This will aid them in achieving sustained mucosal recovery, which holds the potential for improved patient quality of life.

Conflict of interest

Daniel Leffler has affiliation with Takeda Pharmaceuticals but none of the remaining authors have any conflict of interest.

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