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Creation of Experience-based Celiac Benchmarks – The First Step in Pre-Transition Self-Management Assessment

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Abstract

Objectives—Transition planning for children with chronic disease includes the development of independence in many self-management tasks. Conditions that depend on diet have distinct skill sets not well assessed by the traditional transition-readiness tools. There has been literature that describes age-appropriate skill acquisition for diabetes and food allergy patients. However, there are no age-appropriate benchmarks established for celiac disease (CD).

Methods—CD experts (including physician, nurse, dietician, social worker, patient and parent) created a list of celiac-related tasks, which formed the basis of the survey. Patients with CD, and their parents, were recruited from outpatient celiac clinic and support groups, and invited to report the age each task was mastered.

Results—Respondents included 204 patients and 155 parents. Mean age was 12 years (SD4.6) with average of 4 years since diagnosis. The earliest tasks were mastered by a median age of 8 years, such as recognizing GF as gluten-free, eating safely in a shared space and recognizing basic unsafe foods. Describing the effects of eating gluten or explaining celiac disease to a friend or stranger occurred around age 10. Asking about gluten-free preparation in a restaurant, and identifying gluten-free medications or vitamins was mastered around age 12, while tasks involved with safe domestic travel or assessing risk in a job environment occurred between 14 and 16. The interquartile range was about 4 years for each question. No significant difference seen between patient and parent reports.

Conclusions—This novel patient-centered celiac skill list may improve anticipatory guidance and accelerate self-management skills.

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Conflicts of Interest

There are no conflicts of interest for any author.

Keywords

Transition; Celiac Disease; Self-management Skills

Introduction

Celiac disease (CD) is an autoimmune disorder often diagnosed in childhood. Treatment consists of adherence to a strict gluten-free diet, which requires constant vigilance^{1–3}. Self-monitoring and self-management are important tasks for these patients. Expectations around when parents should step back and children should step forward to manage these tasks independently have not been explored for celiac disease. Studies of children with other conditions which are managed by dietary modification, such as diabetes and food allergy, have described the times when parental involvement declines⁴. In this study, we sought to establish a timeline for mastery of tasks related to following a gluten-free diet that is based on patient and parent experience. Our hypothesis is that these tasks are mastered at a far earlier age than is captured by typical transition planning.

Patients with CD have varying experiences with attention to food preparation and consumption. Our secondary hypothesis was that exposure to dietary concerns through diabetes, or food allergies as well as other family members with celiac, would accelerate the age of mastery. We evaluated whether factors such as gender, method of diagnosis, age of diagnosis, exposure to food restrictions in the family or concurrent dietary issues such as allergy influenced the age of mastery. We also explored whether patients and parents had similar ideas about timing. All of this information can be used to create educational tools and develop anticipatory guidance for patients and families with CD.

Methods

A multidisciplinary group of celiac disease experts—including CD dietitians, CD social worker, pediatric gastroenterologists, parents of children with CD, and adults with CD—collaborated to develop a list of tasks specific to CD self-management. Using this list as a guide, parallel surveys for patients with CD and their parents were developed with items related to the age at which the patient mastered a specific task. Children and parents were also asked at what age they thought an “average child with celiac disease” mastered each task. This strategy was employed to mitigate both potential bias regarding self-assessment and the potential confounder of age at diagnosis. Demographic information was also collected, including age, gender, age at CD diagnosis, whether the patient had biopsy-confirmed celiac disease, comorbidities, and family history of CD, diabetes, or food allergies.

Consecutive patients with CD over the age of 4 were recruited in outpatient clinics at Boston Children’s Hospital between March 2016 and January 2017. Included patients were being seen for ongoing care and had been on the gluten-free diet for at least 2 months. Exclusion criteria were recent diagnosis of CD (within 2 months), developmental delay that would preclude answering questions, and inability to understand English. Parent and patient

numbers do not match as some parents had more than one child with celiac, and patients over the age of 18 were not always accompanied by a parent.

Participants were also recruited through an email announcement to the Celiac Support Group at Boston Children's Hospital. All surveys were anonymous and voluntary. Implied consent, through returning the surveys, was explained in the accompanying letter. The parental survey, which implied consent for use of patient's survey responses, was required for patients under the age of 18.

This study was approved by the Boston Children's Hospital Institutional Review Board.

All statistical analyses were performed using Statistical Analysis Systems statistical software package version 9.4 (SAS Institute, Cary, NC, USA). Paired t-test was used to examine the difference in age of mastery reported by patients and their parents. Analysis of covariance was used to test differences in age of mastering a specific skill across gender (male/female), history of diabetes (yes/no), history of food allergy (yes/no), family history of CD (yes/no), diabetes(yes/no), and food allergy (yes/no). Age of CD diagnosis was included as a categorical as a covariate (<12, 12 yrs. old). A p-value of 0.05 was considered significant. We did not conduct multiple comparisons adjustment for P-values, because we are testing one hypothesis and all the outcome variables are inter-correlated, reflecting age of mastering CD-related tasks. Incomplete surveys were included in the analysis, noting the number who answered each question.

Results

Demographics

Responses were received from 204 patients and 155 parents. The mean age at diagnosis was 7.6 and average years since diagnosis was 4 years (Table 1). Almost 75% were female. Comorbid diabetes mellitus or food allergy was uncommon (8% and 13%, respectively), but many had a family member with celiac disease (33%) or food allergy (22%).

Tasks

The earliest tasks were mastered by a median age of 8 years, such as recognizing GF as gluten-free, eating safely in a shared space and recognizing basic unsafe foods (Table 2). Describing the effects of eating gluten or explaining celiac disease to a friend or stranger occurred around age 10. Asking about gluten-free preparation in a restaurant, and identifying gluten-free medications or vitamins was mastered around age 12, while tasks involved with safe domestic travel or assessing risk in a job environment occurred between 14 and 16. The interquartile range was about 4 years for each question.

There was agreement between patient and parents regarding what an "average child with celiac disease" (others) would do. Patients and parents report "others" as mastering most tasks at a slightly earlier age than the age of mastery for the patient, although this difference was not significant. (See Table 3)

Factors

Females seem to master tasks slightly earlier than males, but the difference does not reach statistical significance. Age of diagnosis does impact mastery, in that patients with later diagnoses estimate older ages of skill acquisition. Patients with personal or family history of diabetes tended to estimate older ages, while patients with personal or family history of food allergy or family history of celiac estimated younger ages, compared with food attention naïve patients. However, none of the comparisons reached statistical significance. (See supplemental table 1 and 2)

Discussion

Overall, pediatricians and pediatric specialists are increasingly aware of the need for a gradual transition process from pediatric to adult based health settings⁵. This is true for a wide variety of special health care needs, and gastrointestinal conditions are included. There is a specific consensus that celiac patients need assume responsibility for self-care from their parents^{6,7}. However, the general timelines for transition start around age 12⁸ or 16⁹ and our results show that many tasks for diet- based self-management are mastered at a far younger age. This would make sense as children are involved in meals at preschool, elementary school and social settings such as birthday parties at a very young age, and parents are not always present.

There is literature on two similar conditions that involve dietary modification, food allergies and type 1 diabetes. One survey of pediatric allergists described the age at which they expected patients to recognize symptoms of a reaction, ages 9–11, and when to be responsible for carrying the epinephrine and knowing how to use it, ages 12–14¹⁰. However, a similar study looked at caregiver perspectives and found they expected much younger ages, with recognition around 6–8 years of age and 6–11 for epinephrine administration¹¹. For children with type 1 diabetes, the recommendation is to gradually allow children to take responsibility with insulin injections to be able to do the injections around age 10–12^{12,13}. One study looked at the various components of injection, from mounting the needle to pinching the skin to holding pressure at the site¹⁴. The ability to adjust the insulin dose is noted at 10–12, however, the number of mistakes in insulin calculation does not fall until about age 16.

Many position papers on celiac disease describe the need for a gradual transfer of self-care from child to adolescent to adult patient but describe general tasks such as ‘communication with providers’ or ‘establishing familiarity with the healthcare system’^{6,9}. The tasks inherent in adherence to a gluten-free diet is not further expounded. One celiac support website delineates beginning, intermediate and advanced self-management skills¹⁵. These are described in broad terms. For example, initial skills include “avoid gluten exposure”, “develop a food intake plan” and “establish a partnership with a medical team that includes a nutritionist”. Intermediate skills included “assessment of indeterminate or conflicting information”. There is a paucity of literature with more specific tasks or with the age-appropriate timelines for mastery.

The importance of self-management is compounded by the recognition that adult gastroenterologists may perceive celiac as less important than IBD or colon cancer and thus have limited knowledge to pass on to patients. In one study, dietary counseling for 698 adult celiac patients was considered insufficient for 28% from the physician and 12% from the dietician and qualitative data reported physician lack of knowledge and underestimation of the disease¹⁶. In another study of 168 Canadian children with CD, the gastroenterologist information was considered excellent by only 44%, and the dietician by 36% compared 63% to the celiac website information¹⁷.

Adherence to a gluten-free diet is often not recognized as a complex and multifactorial process which involves far more than whether patients choose to follow the diet or not. Barriers to gluten-free diet adherence include cost, availability particularly outside the home, and knowledge about the gluten-free diet. There are psychological costs as well, with children feeling isolated or different¹⁷. Adolescents with celiac have described the stigma that they feel with this special diet¹⁸. Our survey, which looks at mastery, combines knowledge and social skills required to navigate specific tasks.

Personal factors such as symptoms with exposure, familiarity with dietary cautions, duration of disease and manner of diagnosis might affect the timing of task mastery. We looked at whether exposure to dietary cautions, through family or personal history of allergy, diabetes or celiac, affected the time of mastery. We also looked at duration of disease. It would be very interesting to explore whether symptoms with gluten exposure played a role in earlier mastery, as one might expect with the immediate negative feedback from the physical symptoms. It is possible that the later ages found in the diabetic patients reflected a large number of patients found by asymptomatic screening. However, it is also possible that the focus on diabetic management took precedence over gluten avoidance. This would be interesting to explore further in another study.

Limitations

This is a single center study and thus may have less generalizability. Patients attending a celiac clinic or involved with a celiac support group are likely to be more focused on adherence and might have younger ages of mastery. Larger and more diverse celiac populations should be studied. Despite the anonymity of the survey social desirability bias could skew the results, however, we tried to mitigate with asking about “an average child.” The fact that parents tended to assume “the average child” or “other” could learn faster than the patient, and the general agreement between parent and patient reported ages implies that this desire to please may be a minor factor. We note that report of mastery may not correlate with actual mastery and we did not compare responses with other measures of adherence. We did not calculate a response rate which again limits the generalizability. We did not assess which patients had symptoms with dietary indiscretion, as this may hasten learning. Since patients are diagnosed with celiac at different ages, this study was not designed to specifically tease out the learning curve after diagnosis from age-appropriate developmental mastery. Another study is currently underway in adults to help answer the learning curve question.

Conclusion

This is the first study to carefully examine patient and parent reported timelines for mastery of celiac specific self-management tasks. This data can help inform all key stakeholders in guiding education and expectations to help patients with celiac learn important elements of self-management.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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What is known

Self-management (transition) is important for patients with chronic conditions

Timing of transition assessment is recommended at age 12-16

What is new

Celiac related skill mastery starts early, with median of age 8

Skills are mastered slowly throughout the adolescent years

Parents and patients tend to agree on timing of skill mastery

Novel listing of discrete required skills may help with patient education

Table 1

Celiac survey respondent characteristics

n = number who answered question	Mean (SD)	Affirmative answers (%)
Current age (n=204)	12 yrs. (4.6)	
Age at diagnosis (n=204)	7.6 yrs. (4.1)	
Female gender (n=103)		77 (75%)
Biopsy confirmed (n=161)		144 (89%)
Concurrent diabetes mellitus DM (n=159)		12 (8%)
Concurrent food allergy (n=159)		20 (13%)
Family history of:		
Celiac (n=158)		52 (33%)
Diabetes mellitus (n=156)		5 (3%)
Food allergy (n=154)		30 (22%)

Not all surveys were complete so percentage is based on those who answered in the affirmative divided by the number of respondents who answered that question.

Table 2

Age at which celiac patients reported mastery of self-management tasks

Variable	N	25 percentile	Median in years	75 percentile
Washing hands without the help of an adult	148	3.0	4.0	5
Asking if food was safe to eat	148	5.0	8.0	11
Identifying that certain foods were unsafe to eat	149	6.0	9.0	11
Recognizing GF symbol as Gluten Free	151	6.0	8.0	11
Able to eat safely in shared space	130	6.0	8.0	11
Recognizing and describing effects of eating gluten on your body	142	6.0	9.0	12
Taking vitamins on your own without adult supervision	102	8.0	10.0	12
Able to explain celiac to a friend	144	7.0	10.0	12
Able to explain celiac to an authority member	141	7.0	10.0	12
Able to explain celiac to a stranger	128	8.0	10.0	13
Able to plan your own GF options	119	9.0	11.0	14
Able to identify safe options for food and drink in restaurants	128	8.0	11.0	13
Able to ask about food preparation in restaurants	109	10.0	12.0	14
Able to identify that medications were GF	76	10.0	12.5	14
Able to identify that vitamins and/or supplements are GF	95	10.0	12.0	14
Able to negotiate domestic travel plans regarding your celiac condition	62	10.0	14.0	15
Able to assess risk to their Celiac condition during job opportunities	50	12.0	15.0	16
Able to negotiate travel plans internationally regarding your celiac condition	44	12.5	14.5	16

Table 3
Ages at which patients and parents reported mastery of skills related to following a gluten-free diet

Skill	Patient				Parent report						
	Label	n	Mean	SD	Label	N	Mean	SD	p ¹	p ²	p ³
asking if food is safe to eat	You	129	8.3	3.4	Your child	155	7.6	3.5	0.09	0.03	0.36
	Others	85	7.5	3.1	Others	107	6.7	2.8			
identifying that certain foods are unsafe to eat	You	130	8.6	3.4	Your child	156	7.8	3.4	0.048	0.02	0.03
	Others	83	7.7	2.9	Others	109	6.8	2.7			
recognizing GF symbol as Gluten Free	You	130	8.6	3.2	Your child	149	8.4	3.1	0.19	<0.001	0.07
	Others	83	7.7	2.7	Others	106	7.1	2.4			
able to eat safely in shared space	You	112	8.4	3.3	Your child	125	8	2.9	0.64	0.52	0.53
	Others	76	8.3	2.8	Others	107	7.7	2.7			
recognizing and describing effects of eating gluten on your body	You	123	8.9	3.7	Your child	135	8.5	3.4	0.61	0.01	0.08
	Others	77	8.7	3.5	Others	88	7.4	2.5			
able to explain CD to a friend	You	126	9.6	3.4	Your child	144	8.9	3.1	0.54	<0.001	0.56
	Others	81	9.2	3	Others	103	8	2.6			
able to explain CD to a stranger	You	110	10.3	3.3	Your child	133	9.5	3.1	0.90	0.09	0.28
	Others	72	10.3	2.9	Others	95	8.7	2.7			
able to plan own GF options	You	105	11.1	3.3	Your child	110	10.5	3.2	0.38	0.18	0.71
	Others	72	11.6	3.3	Others	92	10	2.7			
able to identify safe options for food and drink in restaurants	You	112	10.7	3.1	Your child	120	10.5	2.9	0.19	0.06	0.82
	Others	78	10.3	2.9	Others	95	10.1	2.6			
Age when able to ask about food preparation in restaurants	You	96	11.6	3	Your child	92	11.8	3.1	0.41	0.10	0.16
	Others	67	11.4	2.8	Others	93	11.6	2.9			
Age when able to identify that medications were GF	You	66	12.3	2.9	Your child	68	12.6	2.9	0.20	0.39	0.47
	Others	56	13.1	3.4	Others	66	12.8	2.9			
Age when able to identify that vitamins and/or supplements are GF	You	82	11.9	3.1	Your child	87	12	2.9	0.58	0.14	0.60
	Others	61	12.3	3.5	Others	74	11.7	3			
Age when able to negotiate domestic travel plans regarding your celiac condition	You	54	13.1	2.9	Your child	49	13.5	3	0.45	0.25	0.60
	Others	42	13.6	3.4	Others	65	14.6	2.9			

Skill	Patient				Parent report				
	Label	n	Mean	SD	Label	N	Mean	SD	p ¹ p ² p ³
Age when able to assess risk to their Celiac condition during job opportunities	You	42	14	2.8	Your child	43	14.3	2.3	0.33 0.66 0.1
	Others	46	15.6	2.9	Others	64	15.5	2.2	
Age when able to negotiate travel plans internationally regarding your celiac condition	You	39	14.3	3	Your child	32	15.2	2.6	<0.01 0.52 0.1
	Others	40	15.9	3	Others	58	16.8	2.4	

p¹, Comparing self-management of CD between patients with other children with CD, reported by patients

p², Comparing self-management of CD between patients with other children with CD, reported by parents

p³, Comparing patients and their parents, regarding patients self-management of their CD