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ORIGINAL ARTICLE

Impact of a Child's Celiac Disease Diagnosis and Management on the Family

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Abstract

Background Little attention has been paid to family-wide repercussions of a child's celiac disease diagnosis and concomitant gluten-free diet management.

Aims We quantitatively and qualitatively describe positive and negative family-wide effects of a child's celiac disease diagnosis and disease management.

Methods We interviewed 16 families with at least one child currently following a gluten-free diet, with a biopsy-confirmed celiac disease diagnosis ≥ 1 year prior. Mothers and fathers independently rated child's dietary adherence, concern about child's health status, burden in caring for child's dietary needs, and level of change in various aspects of life post- diagnosis. Children rated their own celiac-specific quality of life through a validated scale. Seventy-one in-depth semi-structured interviews were conducted with 16 children with celiac disease, 31 parents, and 24 siblings.

Results Mothers and fathers rated the effects of their child's celiac disease differently, with mothers reporting more lifestyle changes and heavier burden. Negative and positive themes emerged from the interviews. Mothers felt the burden of managing a gluten-free diet. Fathers felt guilty for carrying a celiac disease-associated gene and both fathers and siblings regretted limited food choices at restaurants and home. The need to be a more creative cook was seen as a positive effect by mothers. Fathers appreciated new family traditions. Siblings felt they had developed empathy for others. A framework is proposed to illustrate these family-wide interactions.

Conclusions A child's celiac disease diagnosis and disease management affects the entire family. Our results will inform family-centered interventions that maximize quality of life for families.

Keywords Celiac disease · Family · Qualitative methods · Quality of life · Gluten free diet

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Introduction

Celiac disease (CD), a common immune-based disorder, is characterized by autoantibodies and damage to the lining of the small intestine triggered by dietary gluten [1]. CD affects about 1% of the population, worldwide, with rates higher for first- and second-degree relatives [2, 3]. Symptoms may include diarrhea, anemia, and neuropsychiatric symptoms, or there may be no symptoms at all [4]. If untreated, CD can lead to infertility, osteoporosis, lymphoproliferative malignancy, and other autoimmune diseases [4]. Currently, the only treatment for CD is a strict lifelong gluten-free diet (GFD) which avoids foods containing wheat, barley, and rye. However, dietary adherence is variable as a recent systematic review of studies of dietary adherence in children revealed rates of adherence to a GFD ranged from 23 to 98%, irrespective of the method of determining adherence [5]. More than 40% of patients with CD undergoing follow-up biopsy do not have mucosal healing after a median of 1.3 years indicative of potential ongoing gluten ingestion, which can slow recovery [6].

Managing a diet as restrictive as a GFD presents many challenges as it eliminates foods commonly used in a variety of cuisines and cultures. Once diagnosed, individuals must learn what foods to eat, what foods to avoid, hidden sources of gluten, and how to navigate social situations both in and out of the home. Family involvement is inevitable, especially if the individual diagnosed is a child. Family support is needed not only to ensure strict GFD adherence, but also to support the social and emotional health of the patient with CD.

It is vital to understand how people manage any chronic disease, as complications can have serious consequences. Research has shown the difficulties in adhering to a GFD as well as the financial, social, and emotional burdens it places on participants [7–10]. Qualitative research on CD has captured the burden of illness, quality of life (QOL) issues, and the lived experiences of individuals with CD. A few studies have investigated the management of CD inside the home and more frequently outside of the home to gain a deeper understanding of what the daily management of CD entails [11–18]. Many of these studies have provided insight into how individuals cope with the demands of following a GFD through surveys, interviews, and focus groups. The qualitative research on CD has looked primarily at the *individual* experience of managing CD, with limited attention to the ways families experience a CD diagnosis and GFD (i.e., in terms of household decisions, family food traditions, and negotiating with extended family members) [11, 12, 16, 18].

This study aimed to examine the family-wide effects of CD. Our purpose was to inform future family-based intervention strategies to maximize GFD adherence in patients with CD, while maximizing QOL for all household members. Both qualitative and quantitative methods were used.

Materials and Methods

Design

This was a convergent parallel mixed methods study using a mostly qualitative approach based on phenomenology, which draws upon the lived experiences of interviewees. The study was approved by the Institutional Review Boards of Columbia University Irving Medical Center and Teachers College, Columbia University (# AAAR71700 and #18-222).

Setting and Participants

The study was conducted at the Celiac Disease Center of Columbia University Irving Medical Center (Celiac Disease Center) in New York City. Inclusion criteria required that families have a child aged 8–18 years with a duodenal biopsy-confirmed CD diagnosis ≥ 1 year prior and reportedly following a GFD, and have at least one parent and at least one sibling (with or without CD and between the ages of 8 and 18 years) willing to be interviewed. Exclusion criteria included families with a parent who also had a physician- or biopsy-confirmed CD diagnosis. Families received a $50 gift card for their participation.

Enrollment

Screening, enrollment, and data collection occurred concurrently. The target sample size was 16 families, with "family" defined by the participants themselves. Flyers were posted, and an email blast was sent to all Celiac Disease Center affiliates in the New York Metropolitan area. Out of 5613 emails sent, there was an open rate of 29.9%. (Ninety-five families expressed interest; 69 were screened by phone, 26 did not respond after initial contact. Of the 69 screened, 31 were ineligible due to geographical distance or having an adult with a diagnosis of CD in the household. Among the 38 eligible, 16 were interviewed in their homes. The remaining 22 families were not pursued after screening as the enrollment target had been met and data saturation in terms of family traditions (e.g., Thanksgiving, Christmas, and Passover) had occurred. Some families had multiple age-appropriate children with a CD diagnosis. In that case, the reference child was the one diagnosed first. All interviews were conducted in the homes of the participants by one researcher (CR). Family members were interviewed separately unless they specifically requested

a joint interview. Mothers, fathers, and reference children completed the survey questionnaires after the interviews. Photographs of kitchens were taken during a kitchen tour which was typically led by the reference child.

Data Collection and Measures

Qualitative Measures

The qualitative portion of this study used an interpretative phenomenology approach to examine the lived experience of families [19]. The qualitative data collected enriched the quantitative data by highlighting the complexity of navigating CD and managing a GFD in a family setting.

Interviews

The researchers developed interview questions to capture the overarching research question, "What is the ripple-effect in families following a child's CD diagnosis?" Four main questions were: (1) How do parents describe their experience with their child's CD and managing a GFD? (2) How do children with CD describe their experience with CD and the GFD? (3) How do siblings describe their own QOL, and (4) How do parents, children with CD, and siblings describe family food traditions? Each main question had additional sub-questions probing for specific strategies used to promote GFD adherence and QOL.

Questions were developed with guidance from researchers in the field of celiac disease ($n=5$) and qualitative methods ($n=2$) and pilot tested with families that had children with CD but were not part of the current study ($n=2$). In addition to interview transcripts, field notes were written, with summaries of main points including informal conversations not captured by the audio recordings. The qualitative data analysis used a systematic approach in keeping with best practices in qualitative data methods to code and analyzes the data [19–22]. Once an interview was conducted, it was transcribed in a timely manner by a confidential transcription service. Each transcript was reviewed for accuracy and the interviewer conducted preliminary inductive coding. Transcripts were read through again and coded line-by-line to identify key concepts and ideas. Transcripts were then re-reviewed and coded again to create subcategories. Relevant text was selected from the transcripts based on the research questions and repeated ideas were grouped together as larger themes. Sample responses and/or quotes were selected as illustrative of the themes. The researcher frequently referred to the research questions during the course of the analysis process.

Photographs

The interviewer took photographs of the participants' pantries and kitchens to show how they organized their space, including what special kitchen equipment they used, and gluten-free products. Collecting photographs offered additional insight into how families implement the GFD and potentially creative ways families have chosen to organize their home [23–25].

Quantitative Measures

Demographic and Medical History Variables

Data collected include age of mother, age and gender of all children ages 8–18 living at home, year of the reference child's diagnosis of CD, mother's educational level, household income, and whether other children in the family had CD. Mothers also reported on the reference child's symptoms when exposed to gluten, if the child followed a special diet in addition to the GFD, additional medical conditions, and whether or not a registered dietitian was seen regularly (and if not, why not).

GFD Adherence

Adherence was measured in two ways: (1) Celiac Disease Adherence Test (CDAT), completed by each parent on behalf of their child and (2) The Biagi Adherence questionnaire, completed by each parent on behalf of their CD children and also by the CD children themselves. The CDAT has been validated for adults reporting on their own adherence, but not for adults reporting on their child's adherence [26]. For the purposes of this study, questions were reworded for parents to answer on their child's behalf. Each of the seven items is scored on a 5-point scale, allowing for total scores between 7 and 35. Higher scores indicate lower dietary adherence, scores >13 suggest inadequate adherence. The Biagi four question survey has been used for both adults and children [27, 28]. Scores range from 0 to 4, with lower scores indicating worse adherence, scores ≥ 3 suggest strict GFD adherence. For the purposes of this study, questions on the both scales were reworded for parents to answer on their child's behalf. Additionally, we modified the wording of question 4 on the Biagi scale to read "Does your child only eat packaged foods that have the certified gluten free label?" versus "Do you only eat packaged food guaranteed by the Coeliac Association?" to correspond with U.S. labeling practices. We also added a question to the Biagi scale, "Would your child consume a food that has all gluten-free ingredients with the label 'This Product Was Processed in a

Facility That Processes Wheat'?" (All the time/Sometimes/Never) to capture descriptive information on how children with CD and their parents handle this common situation.

CD-Specific Quality of life

QOL was measured by the Celiac Disease Pediatric Quality of Life (CDPQOL), which has been validated for children ages 8–18 [29]. Children completed either the 13-item questionnaire (ages 8–12 years) or the 17-item questionnaire (ages 13–17 years). Subscales for both the children (e.g., negative emotions, school, and enjoyment) and teenagers (e.g., social, uncertainty, isolation, and limitations) were computed. Total scores and subscales scores range between 0 and 100 scale with higher scores indicating a higher QOL as specified in the Users' Manual [29].

Caregiver Concern

The Ferretti Caregiver Questionnaire was used to examine caregiver concern and impact of CD. Visual analogue scales (VAS) allow quick and reliable assessments of concern [30]. VAS scores ≥ 7 indicate a "high" burden, while scores < 7 indicate "low" burden. For this study, the questionnaire was modified slightly to reduce overlap with the demographic survey instrument (e.g., education level, child's age at diagnosis) and to exclude symptoms not applicable to children (e.g., infertility or recurrent miscarriages). A question was added at the end of the questionnaire: "How much of a burden is caring for your child's dietary needs?" assessed with a VAS scale consistent with the others.

Participants were asked to complete these questionnaires after the semi-structured interviews. Quantitative results were compared to data from the qualitative interviews as a way to triangulate the data.

Data Analysis

Means and standard deviations are presented for continuous data, frequencies and percentages for categorical data. Paired *t* tests were used to assess group differences in caregiver concern, mothers versus fathers and, for the Ferretti, pre- versus post-CD diagnosis and time of CD diagnosis versus now. We considered $p < 0.05$ as statistically significant. Quantitative data was analyzed using SPSS statistical software [31].

Results

Families were interviewed between April and July 2018. Home visits lasted 2–3 h and individual interviews lasted, on average, 30–45 min with adults and 10–30 min with children. In total, there were 74 photographs, 1313 min of recordings and 476 pages of interview transcripts coded. The main interviewer did reach out to 3–4 families following the interview to clarify responses from the questionnaires. Seventy-one interviews were conducted: 16 mothers, 15 fathers, 16 reference children with CD, and 24 siblings (6 with CD and 18 without CD). With one exception, participant families were two-parent, mother/father households. There was one single-parent, mother only household. Overall, families had a high socio-economic status with all self-reporting household annual incomes greater than $100,000, all mothers reporting a college education, and the majority (68.8%) having education beyond college. Fifty percent of mothers were employed (either working from home or outside of the home) and 100% of the fathers were employed outside of the home. The mean (SD) age of the reference children with CD was 12.6 (2.8) years (range 8–17 years) and 56.3% were female; mean (SD) age at diagnosis was 7.6 (3.7) years and children, on average, had lived for 5.1 years with CD (range 1–11 years). By virtue of the inclusion criteria all reference children had at least one sibling. The 24 siblings interviewed were, on average 13.2 (2.7) years old and 50.0% female. Five out of the 16 families had multiple children with CD ($n = 6$ siblings). Ten siblings were older and twelve were younger than the index CD sibling. There were also two families in which one twin had CD and one did not. Of the 16 families, only 3 (18.8%) reported seeing a registered dietitian on a regular basis at the time of the interviews.

Child's GFD Adherence and QOL

In general, mothers, fathers, and children with CD rated their GFD adherence as high on both the CDAT and Biagi questionnaire. Mothers' CDAT mean (SD) 9.1(1.9), Biagi 3.4 (0.5) and fathers' CDAT 9.3 (1.7), Biagi 3.1 (0.7). The differences, mothers versus fathers, were not significant. Children's self-ratings were consistent with those of their parents and also with the qualitative data collected. For the question, "Would your child consume a food that has all gluten-free ingredients with the label 'This Product Was Processed in a Facility That Processes Wheat'?" (All the time/Sometimes/Never), the most common response by mothers and fathers was Sometimes (50% for mothers, 60% fathers), followed by Never (31.3% mothers, 33.3% fathers).

Overall, children and teens scored high on QOL measures, with females scoring higher than males. Scores of the CDPQOL ranged from 69 to 97 out of 100. There was no significant association between years since diagnosis and CDPQOL scores. Children with more severe symptoms when exposed to gluten tended to score lower, $n = 8$ [73.1 (17.8)] than those with no symptoms $n = 6$ [82.1 (5.6)] or milder symptoms $n = 2$ [97.3 (1.7)], however these differences were not statistically significant.

Caregiver Concern

The Ferretti Caregiver Questionnaire scores indicated significant changes pre- to post-diagnosis for both mothers and fathers. On average, mothers scored 4.4 (4.5) when rating their concern before diagnosis, 7.6 (3.0) after diagnosis, and 4.8 (2.4) for concern now. Fathers, on average, scored 4.4 (4.2) when rating their concern before diagnosis, 7.3 (3.0) after diagnosis, and 4.9 (2.9) for concern now. For both mothers and fathers, there were significant changes in scores from before diagnosis and after diagnosis (mothers: $t = -2.9$, $p < 0.05$; fathers: $t = -2.4$, $p < 0.05$), and after diagnosis and now (mothers: $t = 5.9$, $p < 0.001$; fathers: $t = 3.4$, $p < 0.05$). This indicated that both mothers and fathers were most concerned after diagnosis, compared to before and now.

There was no statistical difference between mothers and fathers in terms of level of concern at these three points. There were statistically significant differences between how mothers and fathers rated the impact on lifestyle and social life aspects, with mothers reporting more impact ($p < 0.001$ and $p < 0.005$, respectively). Additionally, mothers also reported more burden when asked about caring for dietary needs than fathers (Table 1). These results were consistent with interview responses by all members of the family who reported that the responsibility of creating a safe environment fell predominantly on mothers.

Family Members' Impact on the Child

The qualitative interviews revealed that the effects of a child's CD diagnosis were not uni-directional, the effects flowing from the reference child with CD but also from each family member back to the reference child. All members were committed to maintaining a safe environment for the child with CD and actively took steps to limit gluten contamination of food in the home, with some of these steps illustrated by the photographs of the home (Fig. 1). Households had varying degrees of how much gluten was allowed in the home ranging from having no gluten whatsoever, some gluten from outside in designated areas, to having shared gluten and gluten-free kitchens. Mothers were seen as the main educator on the GFD and the one who researched new foods and restaurants. Both mothers and fathers promoted a perseverance and possibility mindset for their children with CD, encouraging them to believe anything was possible (e.g., eating out, travelling) as long as they planned ahead and not to limit themselves because of their disease. Fathers also encouraged independence and confidence in their children with CD. Siblings without CD showed support in addition to tolerance and acceptance of the GFD. Siblings with CD, in addition to also following the GFD, provided education and support.

CD Impact on Family Members

The qualitative interviews revealed both positive and negative impacts of CD. Mothers discussed the burden of taking on the majority of food tasks related to the GFD (e.g.; researching safe products and brands, shopping, cooking, and coordinating with outside institutions like schools and camps). However, they also mentioned development of their own culinary skills as a positive impact, acknowledging that

Table 1 Change in mother versus father's level of caregiver concern following a child's celiac disease diagnosis

	Mothers ($n = 15$) Mean (SD)	Fathers ($n = 15$) Mean (SD)	t^{+++}	p
What was your degree of concern about your child's health status:$^{+}$				
Before the diagnosis of celiac disease	4.4 (4.5)	4.4 (4.2)	0.61	0.952
After the diagnosis of celiac disease	7.6 (3.0)	7.3 (3.0)	0.38	0.706
Now	4.8 (2.4)	4.9 (2.9)	−0.10	0.918
How much did the diagnosis of celiac disease of your child affect your:$^{+}$				
Lifestyle	8.0 (2.5)	5.4 (2.5)	4.39	**0.001**
Economic aspect	2.6 (2.5)	1.3 (2.5)	1.42	0.178
Social life	5.3 (3.4)	2.4 (2.0)	3.32	**0.005**
Working life	1.9 (2.9)	1.3 (2.2)	0.60	0.556
Family life	5.1 (3.4)	3.7 (2.8)	1.35	0.198
How much of a burden is caring for your child's dietary needs?$^{++}$	4.8 (2.7)	3.2 (3.0)	2.20	**0.045**

Statistically significant values ($p < 0.05$) are given in bold

$^{+}$Modified Ferretti's Caregiver Concern Visual Analogue Scale 0–10. Higher values suggest higher changes (0 = no change, 10 = highest change)

$^{++}$Modified Ferretti's Caregiver Concern Visual Analogue Scale 0–10. Higher values suggest higher burden (0 = least burden, 10 = most burden)

$^{+++}$Paired t tests, d.f. = 14

Fig. 1 Photographs of Family Kitchens. (upper left). Some families used color-coded systems; for instance, kitchen equipment like pots, pans, cutting boards, knives, and spatulas were red to indicate gluten-free while items used in gluten cooking were not. Photograph 2 (upper right). In one family, any gluten items or leftovers were placed in a bin labeled "Glutenville" in the refrigerator. Photograph 3 (lower left). In some families, gluten-free items were labeled or marked with stickers. Photograph 4 (lower right). Some families had two toasters, which were often different shapes or colors, to distinguish between what was to be used for gluten-free items. Others had separate shelves or drawers dedicated to gluten-free foods

they became more creative cooks, were more focused on cooking from scratch, and used fewer processed ingredients. Fathers also discussed cooking as a culinary challenge, with many finding that cooking gluten-free allowed them to be creative and produce delicious gluten-free meals. A negative for fathers was the limitation of dining options and loss of the spontaneity of being able to go to any restaurant when traveling. Siblings without CD mentioned the development of empathy for others as a positive impact of having a sibling with CD, but also reported as negative the impact of maintaining a safe gluten-free environment and limited choice in restaurants. Siblings with CD reported appreciation for having another sibling with CD because it helped to maintain a safe home. Interestingly, siblings with CD reported that they sometimes approached the GFD differently than the reference child based on symptoms and strictness, and this was seen as a negative impact. See Table 2 for illustrative quotes.

Table 2 Illustrative quotes on the Impact of CD on family members

Family member	Themes	Illustrative quotes
Mothers	*Positive impact on mothers* Improving health outcomes of child with celiac disease Cooking healthier and more creatively *Negative impact on mothers* Increasing effort to shop and prepare meals Increasing expenses Increasing time spent coordinating with outside institutions	"Now we've expanded and gotten a little more creative with the cooking. We've gotten some gluten free cookbooks." "I do try to teach her how to make really cool things so that she can bring to a birthday party, you know, this like beautiful bowl of mousse and everyone loves it and no one knows it is gluten-free or they do because she brought it and her friends are supportive." "I'll make dinner about 1:00 in the afternoon and I'll have all the Tupperware lined up with forks taped at the top so there's no driving to Wendy's, there's no McDonald's, there's no quick anything ever, because it's not an option." "One of the reasons I didn't go back to work was her health and we have the choice to do that and I decided to stay home and support that—support her"
Fathers	*Positive impact* Improving health outcomes of child with celiac disease Creating new traditions *Negative impact* Limiting choice in restaurants Feeling guilt for carrying gene	"We go to many sporting events together, so we've created a new tradition of going to California Pizza Kitchen. It has a very good gluten-free protocol. So whenever we go on a—on a trip we head to the California Pizza Kitchen." "We've definitely made a lot of combinations that we found a lot of gluten-free foods actually tastes good, a lot better than regular food" "We were in some part of the city, we tried to find a gluten-free place where we could have lunch. And it—I think it took us like an hour and a half going from one place to the other, asking questions, checking the gluten-free app—if they have gluten-free food here." "Well, from my own personal perspective as the carrier of the gene, there is obviously going to be some element of 'I feel guilty that I'm the one that gave this to them and that I'm asymptomatic so far.' So I can eat whatever I want obviously at home we keep GF. It makes me feel much more compelled to make sure that they eat well."
Siblings with celiac disease	*Positive impact* Helping to maintain a safe home *Negative impact* Approaching GFD differently depending on symptoms and strictness	"Everybody in my family helps me… [my siblings] also do it so it kind of encourages me to do it." "I wasn't too concerned about it because my brother had it before me and I knew already there were ways to work around it." "Well, I think [my sister] is definitely more of a risk taker than I am. I'm a lot more hesitant and a lot more scared to eat out and try new things just because I really don't like the experience of accidentally eating gluten"

Table 2 (continued)

Family member	Themes	Illustrative quotes
Siblings without celiac disease	*Positive impact* Developing empathy for others *Negative impact* Increasing effort to maintain safe gluten-free household Limiting choice in restaurants Receiving less attention	"And so, it's definitely a good learning experience and even though I don't have celiac disease, it kind of opened my eyes to other people who have different diseases or—and to other people who have these problems daily." "It's really not that bad. I actually like a lot of the gluten-free snacks better than some of the gluten snacks and my mom makes really good food. I'm totally fine it's gluten-free." "When we sometimes get soft ice cream and my sister sometimes, she lets me have a cone, but normally, she wants me to have a cup and if I have a cone, she never lets me get sprinkles which I understand it, but it's kind of annoying." "I was jealous because he got to like speak to the waiters about like not just food"

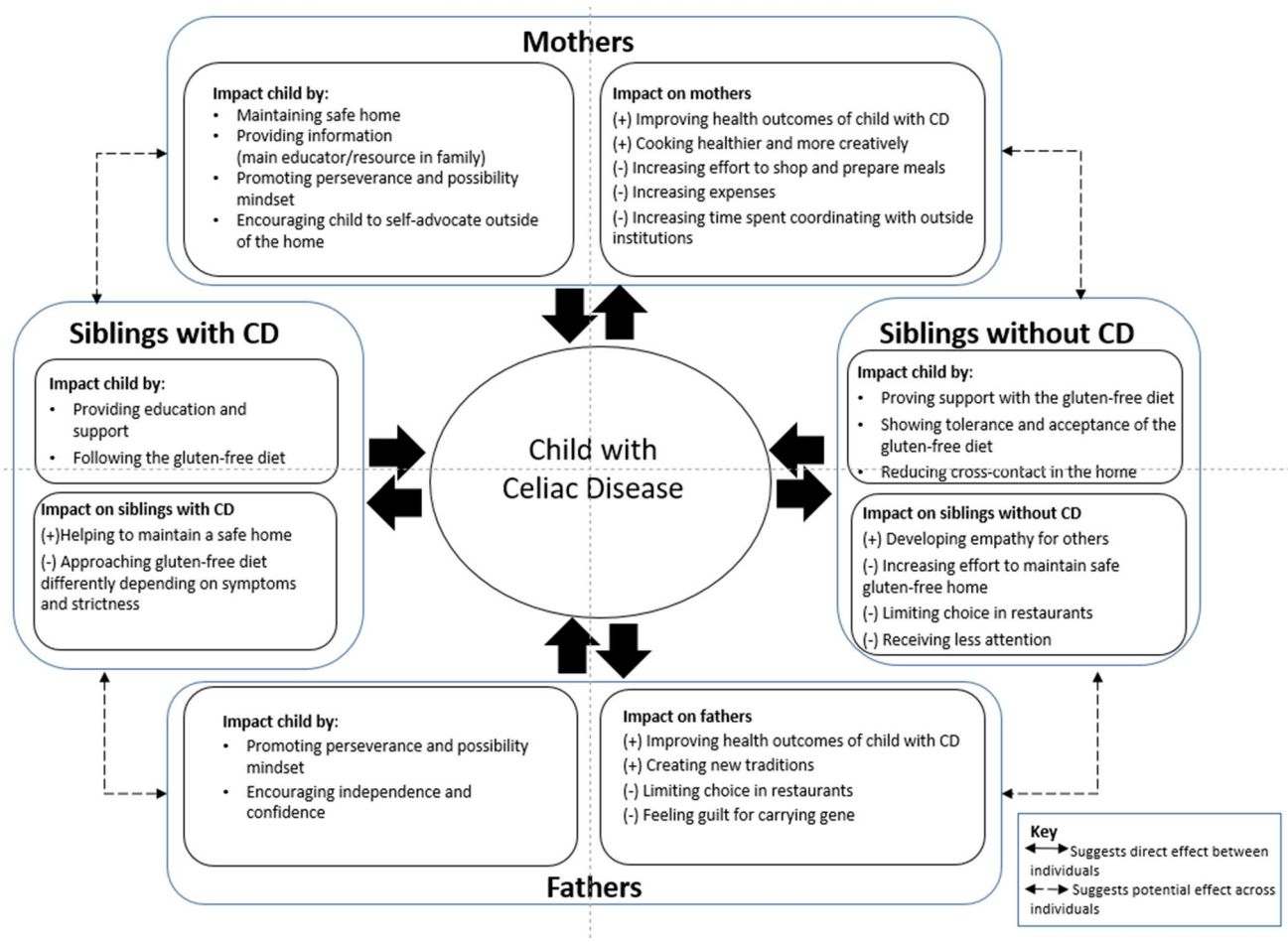

Fig. 2 Family-wide effects of celiac disease

Family Impact Framework

Based on the quantitative and qualitative findings, a bi-directional effect framework was developed that shows potential positive and negatives ways a CD diagnosis can impact family members (see Fig. 2). This research underscores the role family members play in managing CD and GFD management.

Discussion

Overall, the reference children in this study had high levels of self-reported adherence (rated by themselves and their parents) and high levels of QOL (rated by themselves). This was echoed in interviews, which indicated that families generally are successfully navigating the diagnosis of CD and the GFD. This study showed that a child's diagnosis of CD impacts multiple members of the family including mothers, fathers, and siblings, who in turn also impact the child with CD. Mothers tended to report more burden of caring for their child's dietary needs, both qualitatively and quantitatively. Fathers indicated less overall burden than mothers, however were still impacted by CD, for instance, in the limited restaurant options available to them. Siblings were also limited in food choices, whether they had CD or not. Interestingly, family members also discussed positive ways that CD had impacted them, including becoming more creative cooks, developing empathy and appreciating new family food traditions.

A key finding from this study was the development of a bi-directional effect framework that highlights the potential ways the child is impacted and how in turn each family member is impacted by CD. This proposed framework drew upon a framework of diabetes management in a family setting, which suggested that having support in tasks (e.g., food shopping, cooking, picking up prescriptions) from a willing and able family member is crucial in management of diabetes. Assistance in diabetes management, along with encouragement for dietary and exercise habits, were shown to improve glycemic control, which ultimately improved health outcomes [32]. Using themes from the interviews, the ways in which family members supported or impacted the reference child were included in the framework. Having willing and able caregivers and siblings to guide children in management of their CD and the GFD may have potential implications for their health outcomes.

Although many studies have examined the individual experience of having CD, few have looked in-depth into how families as a whole are impacted. This study builds on the current literature, which has demonstrated the impact on relatives of those with celiac disease. One study showed that there was a positive correlation between severity of symptoms reported by participants with celiac disease and the extent of burden their partners reported [16]. The burden and challenges of following a GFD have also been echoed in qualitative studies involving families [11, 12]. However, the majority of the research typically collects data from the parental perspective, most frequently the mother. Family-wide effects and interactions are particularly relevant when a child is diagnosed with CD. This mixed-methods study points to both positive and negative impacts of a child's CD diagnosis on parents and siblings. This ripple effect is important to understand for each family is unique. Healthcare practitioners need to understand that part of the diagnostic process of CD in a child mandates routine follow up which includes ongoing education and support for the family with updates as research is conducted. In fact, a recent study demonstrated that gluten transfer from pots, toasters, and knives may not be as risky as previously thought [33]. Ongoing conversations with families are needed to balance the risk of gluten exposure with the potential harm from hypervigilance and associated anxiety and depression [34]. Our study showed that mothers are predominately the ones maintaining a safe environment for their child in their homes and assume the majority of the responsibility. We have demonstrated that including mothers, fathers and siblings in research studies can lead to a nuanced understanding of what occurs in families.

Furthermore, this study shed light into new areas of research and nutrition education interventions, which include including multiple family members (and possible extended family members such as grandparents).

This study has several limitations. The high socioeconomic status (SES) of study participants limits the generalizability of results, which may not be reflective of other families managing CD. Study families also had high self-reported adherence scores and the children, on average, had high QOL scores, which may have limited the types of experiences discussed in the qualitative interviews. Two measures of adherence were used in this study. Although the CDAT and Biagi questionnaire are both valid instruments, neither has been validated for use by parents on behalf of their child. The Biagi questionnaire has been used with children in a prior study [28]. Furthermore, adherence, caregiver concern, and quality of life were all self-reported measures. Furthermore, this study recruited from the Celiac Disease Center at Columbia University, which excluded the experiences of families who received education and support from other medical centers in other geographic areas. This study benefited from having the researcher in the homes, particularly in having a tour of the kitchen by the family members, which would not have been possible if recruitment did not have geographic limitations.

Conclusions

A child's CD diagnosis and concomitant GFD management affects the entire family. Our results can help inform family-centered CD interventions that promote GFD maintenance while maximizing QOL for all involved. Future research is needed in more diverse families, particularly those families with lower SES status and in other areas of the country.

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Compliance with Ethical Standards

Conflicts of interest CR, HL, RW, PZ, NR declare they have no conflicts of interest. AL is on the Medical Advisory Boards for Dr Schar Foods and the University of Monash Low FODMAP Diet. BL serves as a consultant for Takeda, Innovate, and Anokion. PG consultant to Janssen/J&J, Innovate, ImmunogenX. Speakers bureau—Abbvie.

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

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

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