BMC Gastroenterology

BioMed Central

Research article

The Canadian celiac health survey – the Ottawa chapter pilot

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Published: 11 May 2003

BMC Gastroenterology 2003, **3**:8

Received: 10 February 2003
Accepted: 11 May 2003

This article is available from: http://www.biomedcentral.com/1471-230X/3/8

Abstract

Background: Celiac disease may manifest with a variety of symptoms which can result in delays in diagnosis. Celiac disease is associated with a number of other medical conditions. The last national survey of members of the Canadian Celiac Association (CCA) was in 1989. Our objective was to determine the feasibility of surveying over 5,000 members of the CCA, in addition to obtaining more health related information about celiac disease.

Methods: The Professional Advisory Board of the CCA in collaboration with the University of Ottawa developed a comprehensive questionnaire on celiac disease. The questionnaire was pre-tested and then a pilot survey was conducted on members of the Ottawa Chapter of the CCA using a Modified Dillmans' Total Design method for mail surveys.

Results: We had a 76% response to the first mailout of the questionnaire. The mean age of participants was 55.5 years and the mean age at diagnosis was 45 years. The majority of respondents presented with abdominal pain, diarrhea, fatigue or weight loss. Prior to diagnosis, 30% of respondents consulted four or more family doctors. Thirty seven percent of individuals were told they had either osteoporosis or osteopenia. Regarding the impact of the gluten-free diet (GFD), 45% of individuals reported that they found following a GFD was very or moderately difficult. The quality of life of individuals with celiac disease was comparable to the mean quality of life of Canadians.

Conclusion: On the basis of our results, we concluded that a nationwide survey is feasible and this is in progress. Important concerns included delays in the diagnosis of celiac disease and the awareness of associated medical conditions. Other issues include awareness of celiac disease by health professionals and the impact of the GFD on quality of life. These issues will be addressed further in the national survey.

Background

Celiac disease or gluten sensitive enteropathy (GSE) is a genetically based immune-mediated disease characterized by a life-long intolerance to gluten. In celiac disease the small intestinal mucosa is damaged by the gluten proteins contained in wheat, barley and rye. The damage to the small mucosa results in impaired absorption of iron, calcium, folate and the fat-soluble vitamins A, D, E, and K.

Elimination of gluten from the diet usually results in full mucosal recovery.

Celiac disease may manifest with varying symptoms and severity [1]. The variety of clinical presentations can make the diagnosis of celiac disease challenging, and may result in delays in diagnosis [2]. Celiac disease can express itself as a skin condition referred to as dermatitis herpetiformis (DH). A 1989 survey of the Canadian Celiac Association (CCA) members reported delays in diagnosis ranging from 5.8 years for those with nausea and vomiting to 13.9 years for those individuals presenting with headache. A United States based survey estimated that the mean delay in diagnosis was 11 years after the development of symptoms [3].

Over the last decade there has been an increased awareness that untreated celiac disease is associated with the development of other diseases. Celiac disease is associated with an increased risk of other immune-mediated diseases such as diabetes and there is believed to be a relationship between the length of exposure to gluten and development of other diseases [4]. Continued exposure to gluten in individuals with celiac disease can lead to an increased relative risk of developing small bowel lymphoma [5]. The association between celiac disease and osteoporosis has been widely recognized [6]. Prompt and early diagnosis of celiac disease could potentially prevent the development of a number of these associated diseases.

It is not clear whether celiac disease and the necessity of following a lifelong GFD has an impact on quality of life. In a Swedish study subjects with celiac disease who followed a GFD for ten years did not rate their quality of life as high as age and sex matched members in the general population [7]. There is no current data on the quality of life in Canadian individuals with celiac disease.

The prevalence of celiac disease is estimated at 0.4% of the population in European countries and 0.5% of the United States population [8]. A recent population study from Sweden indicated a prevalence of 1 in 188, but estimates that 8 out of 10 cases have not been diagnosed [9]. In Canada it is estimated that 1 in 250 Canadians may have celiac disease.

Similarly, it is estimated that 10% of first degree relatives of individuals with CD have this condition. First-degree relatives of individuals with celiac disease are also thought to have an increased prevalence of other autoimmune diseases [10].

The last national survey of members (N = 1937) of the CCA was conducted in 1989 [11]. Since that time the membership has grown and there is increased recognition that celiac disease frequently does not present with the classical gastrointestinal symptoms [1]. We wanted to examine the relationship between presentation of symptoms and time to diagnosis. We also wanted to determine what proportion of members and their first-degree relatives had associated medical conditions.

Objectives of the survey

The objectives of the pilot survey were:

1. To determine the feasibility of surveying the full membership of the CCA of over 5,000 members.

2. To obtain more information about length and nature of the diagnostic process.

3. To determine what proportion of individuals with celiac disease and their first-degree relatives had associated medical conditions.

4. To evaluate the quality of life and compare the results to Canadian normative data.

5. To assess what proportion of members adhere to the diet and to identify difficulties with dietary compliance.

Methods

The Professional Advisory Board of the CCA in collaboration with the Department of Epidemiology and Community Medicine of the University of Ottawa developed the questionnaire. The study was funded by the J.A. Campbell Research Fund of the CCA. Two consultants were employed to assist during the initial development of the project and the questionnaire [12]. The questionnaire consisted of eleven different sections, 77 questions and was 15 pages long. The final draft of the survey was reviewed by two international experts in celiac disease. Ethics approval to conduct the pilot survey on Ottawa members was received from the Ottawa Hospital. The survey included questions on demographics, diagnosis: whether diagnosis was confirmed by small bowel/skin biopsy or antibody test, clinical symptoms prior to and after diagnosis, misdiagnoses, delay between onset of symptoms and diagnosis, associated medical conditions of members and first-degree relatives. Quality of life was evaluated with the generic quality of life instrument the SF-12 and there were sections on bone disease and reproduction [13]. Special celiac issues relating to diet were identified including: travel, limitations on social life, access and quality of gluten free food [1].

A pre-test of the survey was conducted by members of the CCA Board of Directors and a formal pre-test was conducted on 14 CCA members (males and females with CD and DH) for readability and ease of completion.

A pilot of the questionnaire was conducted on the 414 members of the Ottawa Chapter of the CCA in October 2001. A coded mailing list of subjects was used to ensure confidentiality and the surveys were mailed by the CCA to all members of the Ottawa Chapter. Only one member per household was instructed to complete the questionnaire. Members were reminded of the survey in the Ottawa Chapter's newsletter and at the monthly meeting. We used a Modified Dillmans' (1978) Total Design method for mail surveys when designing the pilot survey [14].

Data Analysis

Data was analyzed using SPSS v10 for Windows. Logic checks were done using cross-tabulations for key variables. The proportion of respondents choosing different options was calculated for each question.

Results

Return and Response Rates

After the first mailout we had a 76% response (315) so a second mailout was not necessary. For purpose of the analysis, we used data only from adult members who had biopsy-proven celiac disease or dermatitis herpetiformis N = 266 (84%).

Demographics

74% females and 26% males. The mean age of participants was 55.5 years (SD 14.7). 85% of participants were Caucasian and 42% had some postgraduate education. The mean age at diagnosis was 45 years. The mean duration of disease was 10.3 years (SD 10).

Clinical Symptoms Prior to Diagnosis of Celiac Disease

Table 2 lists the proportion of individuals presenting with a variety of clinical symptoms prior to diagnosis of celiac disease. The majority of respondents presented with symptoms of bloating, abdominal pain (74%), diarrhea (71.3%), extreme weakness or fatigue (66.3%) or weight loss (64%).

Previous Diagnoses

Respondents were asked if prior to their diagnosis of celiac disease, were they told that their symptoms were due to other medical conditions (Table 3). Of these, 44% of respondents had been told symptoms were due to anemia, 32% of respondents told due to stress, and 24% told their symptoms were due to an irritable bowel syndrome.

Delay in Diagnosis

Prior to diagnosis, 22% of respondents consulted two or more family doctors and 12% consulted 2 or more gastroenterologists. Thirty percent consulted 4 or more doctors about their symptoms. The median delay in diagnosis after onset of symptoms to diagnosis was one year after history of weight loss, 2 years after onset of symptoms of

Table 1: Questionnaire Summary

Demographics
Age, sex, age of diagnosis
Level of education, family income
Diagnosis
Pre-diagnosis symptoms and duration
Status of recovery from symptoms
Other diagnoses
Possible triggers
Number and types of doctors consulted
Diet
Adherence to gluten-free diet
Difficulty in following diet
Rating of information sources
Difficulty with labelling of foods
Quality of Life
SF12 Health Survey ©
Celiac-specific quality of life questions for adults/children
Bone disease
History of fractures
Bone density status
Osteoporosis treatment
Reproduction
Specific questions for women and men
Celiac disease in family members
Testing of first-degree relatives for CD
Suspected CD in family
Other diseases of members/ first-degree relatives
Gastrointestinal, cancer, autoimmune, blood, endocrine, neurological, musculoskeletal, other

Table 2: Clinical Symptoms Prior to Diagnosis (N = 266)

Clinical Symptom	Proportion of Respondents
Bloating, abdominal pain	73.9%
Diarrhea	71.3%
Extreme weakness or fatigue	66.3%
Weight loss	64.0%
Large pale stools	55.0%
Joint pains	29.0%
Nausea	27.0%

Table 3: Diagnoses Prior to Celiac Disease (N = 266)

Medical Condition	Proportion of Respondents
Anemia	46%
Stress	32%
Irritable Bowel Syndrome	24%
Vitamin Deficiency	17%
Food Allergies	11%
Reflux Esophagitis	14%
Chronic Fatigue Syndrome	8%

nausea and vomiting, 4 years after onset of abdominal pain and bloating and 10 years after onset of symptoms of constipation.

Associated Medical Conditions in Individuals with Celiac Disease

Table 4 lists the proportion of individuals with associated medical conditions. Of all respondents, 57% had iron deficiency anemia, 16% depression, 16% had thyroid disease, 6% had refractory celiac disease and 6% had rheumatoid arthritis. Sixty-two percent of respondents said they had their bone density measured. Twenty-seven percent of respondents had been diagnosed with osteoporosis and 10% with osteopenia. Forty-four percent (n = 116) had a history of a previous fracture and of these individuals, 36% had previously broken their wrist. Five percent were taking calcium and 5% vitamin D after diagnosis of osteoporosis/osteopenia. Thirteen percent were taking an anti-osteoporosis medication such as alendronate, risedronate, HRT or raloxifene.

Table 4: Associated Medical Conditions in Individuals with Celiac Disease (N = 266)

Medical Condition	Proportion of Respondents
Iron Deficiency Anemia	57%
Osteoporosis	27%
Depression	16%
Thyroid Disease	16%
Lactose Intolerance	15%
Refractory Celiac Disease	6%
Rheumatoid Arthritis	6%
Type I Diabetes	5%
IgA Deficiency	1%
Lymphoma	0.7%
Sjogren's Disease	0.4%

Table 5: Associated Medical Conditions in First Degree Relatives

Medical Condition	Proportion with One 1st Degree Relative
Iron Deficiency Anemia	40%
Osteoporosis	31%
B12 Deficiency	25%
Lactose Intolerance	20%
Folate Deficiency	19%
Irritable Bowel Syndrome	18%
Rheumatoid Arthritis	17%
Type 1 Diabetes	12%
Hypothyroidism	11%

First-Degree Relatives with Celiac Disease and Associated Medical Conditions

Ten percent of first-degree relatives were reported to have been diagnosed with celiac disease. Table 5 presents the proportion of individuals that had first degree relatives with associated conditions. Forty percent of respondents had one first degree relative with iron deficiency anemia and 31% percent of respondents had one first degree relative with osteoporosis.

Quality of Life

We used the SF-12 scores to estimate overall quality of life and then compared to normative Canadian data on the SF-36 [15]. The mean summary score for physical function (PCS) was 48.7 and 49.9 for mental function (MCS) (N = 243). These scores were very similar to the summary scores for the Canadian population PCS of 49.7 and MCS of 50.9 [16].

Impact of Gluten-Free Diet

Ninety-seven percent of individuals said that they were instructed to follow a gluten-free diet for life. When asked if they followed a strict gluten-free diet 88% said that they did and the remaining 12% followed a partially gluten-free diet. Seventy-eight percent of individuals said that their health improved a lot after starting a gluten-free diet. Respondents were asked to rate the degree of difficulty they experienced with a variety of aspects of the use of the gluten-free diet and finding gluten-free foods. 45% of individuals reported that they found following a gluten-free diet was very or moderately difficult. 11% of respondents avoided travel all or most of the time because of celiac disease. Fifty percent brought gluten-free foods when they traveled. Twenty three percent avoided restaurants most of the time and 50% avoided restaurants some of the time. Twenty-three percent of respondents found it difficult to find gluten-free food in the stores all or most of the time and 58% found it difficult to find GF food some of the time. With respect to the labeling of gluten-free foods, 20% of respondents found it difficult to determine if foods were GF or not, all or most of the time and 60% found it difficult to determine if foods were gluten-free some of the time.

Sensitivity to Gluten

Sixty-four percent of respondents (N = 169) noted a reaction if they accidentally consumed gluten. Of the symptoms experienced, 79% reported diarrhea, 76% bloating or flatulence, 66% abdominal pain, 37% extreme weakness or fatigue and 27% reported nausea and vomiting. The mean time to onset of symptoms was 6.2 hours.

Quality of Information on GF Foods

The perceived quality of information received from various sources varied (Table 5). Eighty-three percent of indi-

viduals felt that the information supplied by the CCA was excellent in contrast to 35% and 12% who felt that the information received from their gastroenterologist and family doctor respectively was excellent.

Discussion

We had an excellent response to our pilot survey of the Ottawa Chapter despite a long and detailed questionnaire that took over 30 minutes to complete. As a result of the pilot survey we have further streamlined the questionnaire.

We confined our analysis to only those members that had biopsy proven disease.

A number of our results are similar to those seen in previous surveys. Eighty-four percent of respondents had biopsy proven disease which is similar to 75% noted in the US national survey by Green et al. [3]. We found that multiple diagnoses are often considered prior to the diagnosis of celiac disease due to the diversity of clinical presentations, which is similar to findings of the 1989 Canadian national survey [11]. In addition, individuals experiencing symptoms of celiac disease often had long delays in diagnosis, which was noted in results of both the 1989 Canadian survey and US survey of adult celiac disease [3,11]. The delay in diagnosis was reflected by the fact that 30 percent of respondents consulted four or more doctors prior to the diagnosis of celiac. The mean age at diagnosis in our study group was 45 years which may be due to the fact we did not include children in this analysis.

Likewise, anemia, stress and irritable bowel disease were the most frequently reported diagnoses prior to celiac disease, which was the case with the previous national survey [11]. The ratio of female to male respondents (2.8:1) was similar to that observed in previous surveys [3,11].

There is a well-documented association between celiac disease and autoimmune conditions such as auto-immune thyroid disease, Sjögren's syndrome and type I diabetes. The proportion of respondents that reported thyroid disease (16%) and type I diabetes (5%) was similar to that noted in other surveys but the proportion of respondents with auto-immune conditions will be more accurately evaluated in the national survey [10].

In terms of presenting symptoms, diarrhea which is the more typical presentation was reported by 71% of respondents which is lower than that reported in the US survey (85%) [3] but higher than that reported in other surveys [17].

Sixty-four percent of respondents reported a reaction if they accidentally consumed gluten which may be higher than expected.

The quality of life of individuals living with celiac disease was comparable to the mean quality of life for Canadians despite the rigid diet and associated medical conditions. Over 75% of individuals felt that their health improved a lot after starting a gluten-free diet.

The majority of individuals felt that the information received from the CCA and their local chapter was excellent, however information received from other health professionals could be improved.

We surveyed those individuals with celiac disease who were members of the Celiac Association which could have resulted in selection bias. Not all individuals with celiac disease will choose to become or remain members.

One potential limitation of our survey is that we did not have a control group of non-celiac subjects however, the purpose of the survey was to describe the clinical characteristics of the celiac population. Another limitation is that this was a cross-sectional survey and so we did not evaluate quality of life in a longitudinal fashion. The data is self-reported which could result in bias, however we feel that this was less of an issue given the confidentiality measures that were taken. The accuracy of some of the data such as bone density measurement is subject to recall bias. We attempted to minimize social response bias by ensuring confidentiality.

Table 6: Quality of Information Received According by Source

Source	Excellent (%)	Adequate (%)	Inadequate (%)	Not Received (%)
CCA (n = 252)	83	17	-	-
Gastroenterologist (n = 228)	35	35	17	13
Dietitian (n = 224)	29	40	22	9
Family Doctor (n = 235)	12	33	21	34

Conclusions

We concluded that on the basis of the excellent response to the pilot survey a nationwide survey was indeed feasible and is currently being completed and will give us further information of celiac disease in Canada. Delays in diagnosis of celiac disease remain an important issue which needs to be addressed given the prevalence of celiac disease [18]. Awareness and appropriate follow-up of associated medical conditions such as osteoporosis and thyroid disease is essential. Screening of first degree relatives for celiac disease is another important issue. Improved training of health professionals including family physicians and nutritionists about celiac disease is also an important area that requires further attention. The above mentioned issues will be appropriately addressed at a population level by the national survey.

Competing interests

None declared.

Authors' contributions

AC participated in the design of the survey, questionnaire, data analysis, and wrote the manuscript. MZ participated in the design and coordination of the survey, was responsible for development of the questionnaire and provided feedback on the manuscript. IG participated in the design of the survey and questionnaire, assisted with data analysis and provided feedback on the manuscript. CS participated in the development of the questionnaire, and provided input on survey design and the manuscript. All authors read and approved the final manuscript.

Acknowledgements

We would like to thank the Professional Advisory Board of the CCA, the members of the Ottawa Chapter of the CCA; the consultants Judy Lawn and Dr. Dan Harvey; Monica Prince for her assistance with the data entry; and Drs. Alessio Fasano, Peter Green and Kent Stobart for reviewing the questionnaire. We received funding for this project from the Dr. J. Alex Campbell Research Fund. Dr. Cranney was funded by an Arthritis Society scholarship and Dr. Graham is a CIHR New Investigator.

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Pre-publication history

The pre-publication history for this paper can be accessed here:

http://www.biomedcentral.com/1471-230X/3/8/prepub