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Title	Dietary gluten avoidance in Canada: a cross sectional study using survey data
Authors	Adriana Mudryj PhD, Anne Waugh BSc, Joyce Slater RD PhD, Donald Duerksen MD, Charles N. Bernstein MD, Natalie Riediger PhD
Reviewer 1	Hasanain Ghazi
Institution	Community Medicine, Management and Science University, Selangor Darul Ehsan, Malaysia
General comments (author response in bold)	<p>1. title: to using, i think only put using without the (to) 2. abstract need to revise background and remove points, results need to include numbers interpretation: why only Ontario mentioned?</p> <p>Thank you for your review and comments. We have revised the title, which no longer includes “using”. The title is now, “Dietary gluten avoidance in Canada: results from the cross-sectional 2015 Canadian Community Health Survey”.</p> <p>We have revised the abstract background to remove mention of gluten-related conditions and clarified the study purpose.</p> <p>The interpretation of the abstract no longer mentions Ontario.</p>
Reviewer 2	Jo Welch
Institution	School of Health and Human Performance, Dalhousie University, Halifax, NS
General comments (author response in bold)	<p>The authors analyzed CCHS data to determine the proportion of Canadians who consume gluten-free food, and where food is produced and consumed by those who do and do not consume GF food. Although they were limited by no information as to the reason for GF consumption, they drew reasonable and clear conclusions. This is an excellent manuscript.</p> <p>None of my very minor points have to be corrected but they would increase readability.</p> <p>Thank you for acknowledging that you felt our manuscript was excellent. We have addressed your minor revisions in the manuscript.</p> <p>line 76: inclusion of "other" here seems odd. Unfortunately, this is how Statistics Canada has categorized locations and we are unable to revise.</p> <p>line 155: This is an awkward partial sentence. We have deleted this sentence.</p> <p>line 175: This sentence lost me, even after 3 reads. This sentence has been deleted for space. It is not a limitation per se, rather a difference in how dietary data can be reported.</p> <p>Some of the journal titles are in caps (2, 7, 28, 39) while all the others are in lower case. Dietetics is misspelled in reference #7. Thank you for pointing out these errors. We have reviewed and revised the reference list to ensure consistency with respect to the journal requirements.</p>
Reviewer 3	Frank Gavin
Institution	
General comments (author response in bold)	Gluten-free diets (GFDs) appear to have been on the minds of increasing numbers of people in recent years. One sign of this is the pronounced increase in the

bold)

number and variety of gluten-free food items available in supermarkets and restaurants. And, as the paper under review indicates, we hear of more and more people who do not have celiac disease following a GFD for one reason or another.

So the paper is topical, but that may make its reliance on results of the 2015 Canadian Community Health Study, Nutrition Survey a problem. It may well be, for instance, that the much greater availability of gluten-free items in restaurants along with the efforts made by restaurants to draw attention to these items has rendered the results related to the location of where those on GFDs consume their meals and snacks somewhat--maybe quite a bit--out of date. The same applies to the percentage of people following a GFD for one reason or another. If it was 1.9% in 2015, it could be a fair bit higher now.

Thank you for your thorough review. This is true, the prevalence of dietary gluten avoidance is likely to increase, rather than decrease. Unfortunately, the data from the 2015 survey was only released in late 2017, and there are no more recent population-based data on dietary gluten avoidance available in Canada. Unfortunately, there is a considerable time lag between data availability, application for and notification of funding, data analysis, and manuscript preparation. Furthermore, we have not seen evidence that accessibility of gluten-free foods has increased substantially in Canada within the last 5 years.

A more fundamental concern is the research questions. The first—what are the demographic and socioeconomic characteristics of Canadians who adhere to a GFD?—is clear, but there is no explanation for why it is important. Perhaps its importance was assumed. The second—what is the location of food preparation and consumption for those who follow a GFD and what are the differences between those following a GFD, those who report no dietary avoidances, and those reporting other, non-gluten, dietary avoidances?—looks to me like two questions.

We have revised the introduction to indicate the value of describing dietary gluten avoidance in Canada. You are correct in that the second objective includes two parts. We have also revised our description of the objectives to improve clarity.

The demographic categories used in the survey make it difficult to answer that first question in sufficient detail. For instance, participants could identify as “white,” “Indigenous,” or “racialized,” but surely each of these categories needs to be broken down if the results are to be meaningful for many readers, patients or clinicians. People of Chinese and west African backgrounds may well have very different diets and different types of food intolerances. The same might apply to Inuit and members of First Nations. There are also just three age categories (2-17, 18-49, and over 50). Many readers would likely be interested in results for narrower age ranges.

Unfortunately, given the small number of Canadians who avoid dietary gluten who completed the survey, we are unable to provide estimates of reasonable quality if broken down further by racial/ethnic groups, or smaller age groups. We reported data in the most granular form we were able to.

As for the second question—or at least the second part of it, which looks to me like a third question—the authors were unable to answer it because “the survey did not

allow for determination of the reason for gluten avoidance” (p. 10) and did not allow the authors to “determine the proportion of the sample who avoid dietary gluten due to CD, non-celiac gluten sensitivity, wheat allergy, or other reasons” (p.11). These are major limitations.

Our third objective (previously described as our second objective), was to describe the eating location and location of food preparation among Canadians who avoid dietary gluten, and compare to Canadians who reported no dietary avoidances, or report dietary avoidances other than gluten. This question was not dependent on reason for dietary avoidance. However, you are correct in that the lack of data regarding the reason for dietary gluten avoidance is a major limitation, which we have noted on page 10, lines 149-150, and again in the limitations section on page 12, lines 192-194.

The authors say the study employed a “patient-oriented approach” and list one person identified as a “patient partner” as an author (someone who is also affiliated with or a member of the same department at the University of Manitoba as three of the other authors). They also name another patient partner who “informed the study” and mention that they heard in the course of their engagement with patients that the relatively low percentage of people in Ontario who follow a GFD may be a result of the cost of the test for celiac disease not being covered by the province. (It’s covered in all other jurisdictions.) The fact that the percentage of people in Quebec who follow a GFD is exactly the same as the percentage in Ontario, however, makes the explanation for Ontario look quite speculative.

Our explanation for a lower prevalence of dietary gluten avoidance in Ontario is speculative. We are unsure of why there are significant regional differences, but the purpose of the discussion section is to provide our interpretation. When we presented these results to the Canadian Celiac Association, their interpretation included the limited access to testing. We could not think of any other reason why Ontario or Quebec would have a significantly lower prevalence of dietary gluten avoidance. We suspected that different regional patterns of immigration may be impacting this significant pattern given some reported differences in CD according to ethnicity. However, racialization was not significantly associated with dietary gluten avoidance in the multiple logistic regression model, which may have supported regional differences in patterns of immigration contributing to difference in dietary gluten avoidance. Our communication with multiple provincial chapters of the Canadian Celiac Association, specifically in Quebec (including French and English chapters), did not indicate any differences between provinces in terms of dietary culture related to dietary gluten avoidance.

The larger question is what exactly it was that made the authors’ approach truly patient-oriented. We are told that members of the Manitoba chapter of the Canadian Celiac Association “contributed to the refinement of research questions for the broader study” but not what that refinement was. How did the questions change as a result of their engagement? Similarly, there are no details about what Anne Waugh (the patient-partner co-author) contributed to the “analysis/interpretation of the data” or how Linda Diffey (the other named patient) “informed the study.” Such details are needed by readers who want to know what

actual differences patient-partners made to a study.

We have removed the patient-oriented approach completely from the manuscript as described in our response to the editors. Ms. Waugh plans to publish a blog or OpEd in response to the publication of this paper to provide a patient voice as to the importance of the study and the interpretation of the findings.

Certainly, the paper would be a hard read for many patients interested in the topic—and probably for many clinicians too. What, for instance, are the “dining constraints” that “can be difficult when eating glute-free in Canada” and what exactly is “the Canadian food system” that may or may not be adequate in responding to dietary needs? Will most readers know what “post-estimation commands” are or what “coarsened exact matching” and “endogeneity” mean? (All three terms, unfamiliar to me despite over six years spent reading trial protocols and reports as a member of the Canadian Drug Expert Committee, can be found on page 8.) A stray fragment, “Though at present, undiagnosed CD remains common,” sits in the middle of a paragraph on page 10, seemingly unconnected to the sentences before and after it. And very near the end of the paper the authors say the study’s results “may also provide credibility to Canadians who follow a GFD.” Nothing earlier in the paper suggests these Canadians lack or need “credibility.” Perhaps the authors simply chose the wrong word. I could go on with further examples.

Our patient partner and co-author, Anne Waugh, has prepared a blog, or OpEd, that we plan to publish in concert with this manuscript that will be more amenable to the general public and truly allows the patient voice to be heard in a way that is not constrained by text limitations and the research publication process and its often inaccessible language, as you have rightfully noted.

We have removed the term ‘credibility’. Our patient partners consistently have their credibility of their need to eat gluten-free questioned, and that was our reasoning for including that term, though it is now removed.

I may not be the ideal patient reviewer for this paper since I have never followed a GFD. A close family member who follows a FODMAP diet and a close friend who has CD, however, have been on GFDs, so I have some awareness of the challenges involved and what has changed over time. This paper, unfortunately, doesn’t reveal or throw into particularly clarifying light anything that wasn’t already known or strongly suspected, e.g. more women than men in Canada follow GFDs and people who follow GFDs have tended to eat at home more than other people. The considerable limitations of the 2015 survey data are very much the equally considerable. limitations of the paper.

Thank you for your review. Unfortunately, data on dietary gluten avoidance has not been available on a representative sample of the Canadian population until this survey was released. Data to investigate time periods trends in Canada are not available. It is our hope to examine changes in dietary gluten avoidance in Canada when the next dietary survey is completed by Statistics Canada, assuming questions pertaining to dietary gluten avoidance are collected. Though this will likely not be collected for some time. Given the magnitude of work involved in collecting dietary data and preparing the dataset for analysis, there is a considerable time lag involved in analysing the data and preparing a manuscript for publication

	that the public may not fully appreciate.
Reviewer 4	Christine Stobart
Institution	Saskatchewan Centre for Patient-Oriented Research, Saskatoon, Sask.
General comments (author response in bold)	<p>This cross-sectional study seeks to characterize Canadians who adhere to gluten free diets, plus identify where these adherents prepare and consume food in comparison to those Canadians who do not adhere to a gluten free diet. It is conducted as patient-oriented research.</p> <p>It is very encouraging to see the authors embraced a patient-oriented approach for a cross-sectional study. It is important to dispel misconceptions and recognize it is the patient voice, not the methodology, which identifies research as patient-oriented.</p> <p>Thank you for acknowledging that we have embraced the approach. We found the process enlightening and were surprised ourselves at how much we learned from patients, and also how it influenced our interpretation. Our patient partner, Anne Waugh, also found the process beneficial. Unfortunately, due to constraints in the manuscript word limit and our collective discomfort with recommendations regarding patient-oriented research, we have opted to remove all mentions of this study being patient-oriented.</p> <p>1. Patient-oriented research The abstract and p. 3 line 26/27 indicate patients were engaged as full partners. This is commendable so expounding on the following further serves to advance the patient-oriented approach: The Canadian Institutes of Health Research definition of patient oriented research indicates patients—those with lived experience--identify the research priority (CIHR, SPOR 2019; cihr-irsc.gc.ca). Would you be able to describe the process used to identify the topic of this study as a patient priority? As previously described, we have opted to re-submit that manuscript not including the POR template.</p> <p>As co-investigators patient partners inform the research process in patient-oriented research. It was useful to hear patient partners were able to aid in data interpretation and provide insight on Ontario's lack of serological testing for Celiac Disease. It would be advantageous to hear what other specific insights patient partners provided such as those of Linda Diffey (p. 2, line 5/6), which the authors found helpful. Delineating these contributions highlights the merit of patient engagement within healthcare research. Specifically, Linda aided in our interpretation of differences in energy intake attributed to various locations. However, as previously described we have opted not to re-submit as a POR manuscript.</p> <p>Integrated knowledge translation emphasizes the important role knowledge users, and notably patients partners in this study, play is the dissemination of findings. Acknowledgements indicate patient partners were involved in knowledge translation beyond this article. Elaboration on their knowledge translation role within the community would be a welcome addition to this manuscript. Unfortunately, this is outside of the scope of this manuscript. As part of our knowledge translation. Dr. Riediger and Mudryj, and patient partner Anne Waugh attended a gluten-free potluck with approximately 150 members of the Manitoba Chapter of the Canadian Celiac Association and presented our</p>

results. Dr. Mudryj will also present at the Canadian Celiac Association (CCA) annual conference. Ms. Anne Waugh prepared an infographic for the CCA for use in their own public engagement, and she has also prepared a blog/OpEd that we hope to publish in conjunction with this publication to raise awareness of the challenges of eating gluten-free in Canada.

Were patients partners compensated for their contributions as co-investigators? Did they provide feedback on their involvement in this study? It is worth noting how patient partner engagement was formally acknowledged, and whether or not patient partners found their involvement meaningful. Providing this information is useful to readers considering the patient-oriented approach.

We had limited funding from the Canadian Celiac Association to complete this study, as such we were unable to compensate Ms. Diffey, who provided insight based on her own interest in the study. Ms. Waugh joined as a patient partner during the course of the analysis. She was an undergraduate student in the Department of Food and Human Nutritional Sciences and seeking a summer undergraduate research experience. Dr. Riediger recruited Ms. Waugh to join the project as a paid patient partner and undergraduate summer student. As such, she was compensated \$7,000 for her work on project, as well as one other project (which she was not a patient partner on). She has embraced this role.

We had no intention of originally submitting this manuscript to CMAJ as we were preparing it. It was actually our engagement with the Canadian Celiac Association, which contributed to our interpretation that the findings had considerable impact. When we noted that CMAJ Open had a POR section, Drs. Mudryj, Riediger, and Ms. Waugh (patient partner) agreed that we thought our study was highly engaged and research questions (on a different manuscript) and interpretation in this manuscript changed as a result of the engagement with CCA. So we reformatted our manuscript to submit as a POR contribution. Ultimately, however, we decided against submitting as a formal POR manuscript during the revision process as a result of concerns from the editor and reviewers, text constraints, and, somewhat ironically, our patient partners wishes. Ms. Waugh plans on submitting an associated blog or OpEd for media distribution, to provide a patient perspective on this manuscript that may provide more flexibility in terms of what she wishes to contribute and to maintain autonomy over the patient perspective. In fact, we would recommend to CMAJ Open, and other journals, to consider an option of having patient partners provide an associated commentary or blog to original research articles.

Were patients partners considered for authorship? Current literature on this topic in CMAJ Open is worth considering should patient partners, in particular AW as a manuscript writer, be amenable.

Her inclusion as an author was more organic rather than intentional.

The GRIPP2 (Staniszewska et al. Research Involvement and Engagement (2017) 3:13 DOI 10.1186/s40900-017-0062-2) is a helpful reporting checklist for patient-oriented research. It is an invaluable guide when describing patient engagement, and would aid in clarifying the role patient partners had within this study.

As discussed with all study authors, we have opted to remove all components of the manuscript related to patient engagement.

2. Cross-sectional study

Although researchers were utilizing the 2015 Canadian Community Health Nutrition Survey which dichotomized sex as male and female, this no longer addresses gender diversity. Results, then, that suggest women had two times higher odds of reporting a GFD compared to men, may not be as relevant as intended. Similarly, a questionnaire based on 24 hour dietary recollection presents recall bias. Both limitations are worth noting.

We have now included the limitation regarding the data collection not addressing gender diversity and recall bias.

3. Discussion

The exclusion of people from the territories, on reserves, and settlements is rightly defined as a significant limitation. Ongoing research in Saskatchewan indicates Indigenous peoples suffer from IBD, and yet rural and remote communities often have limited grocery and food options. As such, this cross-sectional study further highlights the research gap addressing the health and well-being of Indigenous persons in Canada.

We have elaborated on the exclusion of a on-reserve First Nations people and how that contributed to little knowledge regarding the impact of dietary exclusions, particularly gluten, among Indigenous people in Canada. We also heard this concern in our engagement with the Manitoba Chapter of the Canadian Celiac Association.

4. Conclusion

Although succinct, this section ends abruptly. Given this study is patient-oriented, commenting on the extent to which patient partners influenced the overall study outcomes would provide a complete conclusion.

The conclusion has been revised to omit any mention of the study being patient-oriented, and highlighted areas of future research that would further this area of research. We have removed the content about testing. This was insight we gained from our engagement and was considered a priority for the Canadian Celiac Association, but unfortunately, our study was not designed to test for regional differences in celiac disease. Given the challenges with that conclusion as identified by the editors as well as limits with the texts, and in applying a patient-oriented approach, we have removed any text about using a patient-oriented approach.