Author's response to reviews

Title: Technology assessment and resource allocation for predictive genetic testing: A study of the perspectives of Canadian genetic health care providers

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Author's response to reviews: see over
We would like to thank-you for the opportunity to resubmit. The reviewer’s comments were most helpful and have served to strengthen our article. Our responses to the reviewer’s comments are summarized below. The comments from the reviewers are provided in the numbered bolded text below, which are followed by our responses on a point by point basis. These changes are also noted in the revised manuscript.

**Reviewer #1:**

**Discretionary Revisions:**

1. P. 5 para 1: Should “These interviewers” read “These interviews”
   ○ This correction has been made

2. P18: the word “improved” is still in place (should this be approved?)
   ○ This correction has been made

**Reviewer #2:**

No revisions required

**Reviewer #3:**

**Major:**

1. I am still not convinced that the abstract is clear to the readers. In the background section “assessment procedures” were mentioned and in my opinion the authors mean decision rules or guidelines.
The authors have incorporated the reviewer’s suggested change in terminology. The below is included in the abstract section of the paper.

- With a growing number of genetic tests becoming available to the health and consumer markets, genetic health care providers in Canada are faced with the challenge of developing robust decision rules or guidelines to allocate a finite number of public resources.

2. The presentation of the results is still narrative and a clear hypotheses or research focus is missing. I am not sure whether it is possible to condense the material to narrower but clearer meaning. One sentence in the results section does already point to this problem: “They also rated and identified SEVERAL FACTORS that influenced allocation decisions and patients decisions regarding testing.” Unfortunately, this sentence does not contain any specific information which might be due to the fact that the results itself are broad and unspecific.

- The authors feel that the above comments have been adequately addressed both within the abstract and body of the paper.

**Clear hypothesis (as included in the background section of the paper)**

- These interviewers were designed to provide key information regarding the allocation of resources for predictive genetic testing and associated ethical, legal and social issues. More specifically, they explored existing provincial funding structures, criteria and factors that influence resource allocation decisions, relationships between primary health providers and government representatives, the role the media plays in influencing patients requests for genetic tests, and the strengths and weaknesses of the current regulatory framework within which these facilities operate. Policy recommendations regarding changes to resource allocation models and the viability of a national evaluation process for predictive genetic tests were also examined. Information obtained from these interviews have been used to inform ongoing research investigating resource allocation of emerging genomic technologies, the end goal of which is to formulate a list of funding policy recommendations to assist in resource allocation decisions.

**Presentation of results:**

- Due to the small sample size, the results presented are simply meant to describe the findings of the study.

**Rated and identified several factors that influenced allocation decisions and patients decisions regarding testing.**

- The authors feel that this comment has been adequately addressed in the above paragraph, in addition to being more flushed out in the results section of the paper.

- i.e.: Participants were also asked to rate on a scale of 1-5, with 5 being very important and 1 not important, a list of potential factors influencing resource allocation decisions for predictive genetic testing at their facility. These included: cost effectiveness, length of wait times, access to the appropriate equipment, access to new technologies, evidential basis, availability of preventative strategies, ethical/legal consideration, and
media coverage. Participant responses varied, but for each of the factors listed, over half of the participants gave a rating of 3 or higher, with evidential basis and cost effectiveness consistently receiving the highest ratings. Such ratings suggest that all the above factors influence resource allocation decisions for predictive genetic testing in Canada at the laboratory level.

- And
- The pilot study also investigated genetics health care providers’ perceptions of social factors influencing patient decisions about predictive genetic testing. Respondents were asked to rate the influence of patients’ concern about genetic discrimination, impact on family members, ability to obtain insurance, and additional costs associated with a positive result, on decisions regarding testing [17-19]. Providers offered a range of perceptions on patient’s influences. In particular, genetic discrimination was viewed as both, e.g., “A high concern for most patients” and something that “[does not come] up all too often, in our health care system.” Overall, half of the participants (8/16) rated the influence of patients’ concern about genetic discrimination on a scale of 1 to 5, with 5 being very important and 1 not important, as a 3 or higher. Interestingly, a couple respondents (2/16) also commented that factors such as genetic discrimination and the ability to obtain insurance are not normally influential until raised by health providers:

3. A pilot study is generally an empirical investigation to test whether the methods for a main study are appropriate or to test the feasibility of an empirical approach. The presented results are based on a small sample which does not mean that this is pilot study at all, since the main study has different goals and uses a different approach.
   - The authors agree with the reviewer’s observation. The word pilot has been removed.

4. The discussion section is unrelated to the results of the study. Also, the conclusion section goes far beyond the empirical evidence found in the data. Sometimes it reads more like a political statement than an empirical investigation. The authors should have a more neutral position in the interpretation of the “results”.
   - No changes have been made. The authors disagree that the discussion section is unrelated to the results of the study and that the conclusion goes beyond empirical data found.

5. The study was funded by Genome Canada which might have interfered with the results and their interpretation by the authors. Could you please give to the reviewers some information about this organization? Furthermore, it would be interesting to know whether the results reported by the authors are in agreement with the position of the funding organization or not. Another possible threat to the data could be a self selection bias of the interviewees, based on the motivation not to take part in this study due to the funding organization. Could you please
state how the workshop in Banff was announced and whether the participants were informed about the funding organisation.

- **Information on Genome Canada:** Genome Canada is a not-for-profit organization established in February 2000. It was given a mandate by the Government of Canada to develop and implement a national strategy for supporting large-scale genomics and proteomics research projects, for the benefit of all Canadians.

- Funding is awarded through a peer-reviewed process. Currently Genome Canada helps fund 6 Genome Centers: Genome British Columbia, Genome Alberta, Genome Prairie, Ontario Genomics Institute, Genome Quebec, and Genome Atlantic.

- When participants were first contact they were informed that the study was funded by Genome Canada via Genome Alberta. Of those that declined to take part in the study – which was three in total -- none indicated that it was due to the source of funding.

- None of the interviewed participants were involved in the Banff workshop. This workshop was by invitation only, and all participants knew that Genome Canada (via Genome Alberta) was one of the workshop funders. The others included: Genome Atlantic, Genome British Columbia, Alberta Law Foundation, Advanced Food and Materials Network (AFMNet) and GeneSens.

6. **Abstract: the naming of the states is not necessary.**
   - The authors have left the names of the provinces in this section they indicate which provinces were represented in the study.

7. **Abstract: please mention the method of analysis.**
   - The following sentence has been included in the abstract section of the paper.
   - *Given the community sampled was identified as being relatively small and challenging to access, purposive sampling coupled with snowball sampling methodologies were utilized.*

8. **Abstract: What is meant with “a national evaluation process for predictive testing”?**
   - Given that health care delivery within Canada is managed both provincially and federally, creating coordinated health policy is always a concern. While still being sensitive to regional differences, the authors support the idea that having national guidelines and standards, regardless of the technology, is a good place to start.

9. **Page 4: what is the message of the first sentence in the second paragraph? What could be the value? The Discussion gives some information on that.**
   - This sentence introduces the idea that health providers are involved in the decision making process with regards to the utilization of genetic technologies,
which is further discussed in the paper. The authors believe this paragraph is important to the background section of the paper.

10. Page 5: some details about the methods were described and should be removed to the methods section.
   o The authors have left the section as is. The paragraph at the bottom of page 5 they believe the reviewer is referring to provides important background information.

11. Page 7: “all interviews were conducted by the same individuals”. Does this mean that several persons did the interviews? How many interviewers were involved?
   o Two individuals were involved in the interview process. One that conducted the interview, while the other transcribed. Their roles did not change throughout the study.

12. Is there any information about interview quality?
   o By utilizing two individuals -- both of whom are experienced at conducting interviews – and by recording and transcribing open-ended questions, the authors felt that the quality of interviews conducted was high. In addition, questions were asked in the same order, and efforts were used to ensure that the same wording was used consistently.

13. Page 8: BRCA1 instead of BRAC1
   o This error has been corrected.

14. Discussion: were any of the results unexpected?
   o The authors found the answers to the question “Assuming patient consent, who makes the decision about whether a predictive genetic test will be done?” surprising. The majority of participants indicated that more than one professional is involved in this process. The authors were not expecting such variability.