Author's response to reviews

Title: Patient and primary care provider experience using a family health history collection, risk stratification, and clinical decision support tool: A Type 2 hybrid controlled implementation-effectiveness trial.

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Editorial Review Board  
*BMC Family Practice*

Dear Esteemed Editors:

We appreciate your review of our article, “Patient and primary care provider experience using a family health history collection, risk stratification, and clinical decision support tool: A Type 2 hybrid controlled implementation-effectiveness trial.” We have sought to address the reviewers’ comments below and within the manuscript.

Editorial comments:

1. Please state in your revised manuscript the name of the ethics committee that approved your study.

We state that the study was IRB approved at all 3 institutions in paragraph 1, page 6. Please let us know if you need further clarification of this.

Reviewer 1:

1. Overlap in Intro, Methods, and Results with the group’s previously-published paper in BMC.

There should be some expected overlap between the two papers as the previous publication was a report of the methods for this study. We were careful to make sure there is no exact duplication of wording; however, it is expected that there will be many similar thoughts and ideas regarding background, etc.

2. The Title, Abstract and Methods say that this was a “controlled” implementation-effectiveness study, and mention a 3rd primary care clinic where MeTree was not used, stating that it provides a “concurrent control” group. Yet...the rest of the results do not appear to include any data from control subjects nor clinicians. therefore, in this MS, mention of a controlled trial should be deleted (except possibly to say that you are reporting on a subset of results from a larger trial) and the purpose of the CURRENT analyses should be better-described (i.e., this is a report of the uptake and other process measures, and the acceptability of MeTree for clinicians and users in the two practices in which it was implemented. These analyses compared certain process and satisfaction measures according to education, age, size of pedigree, and whether the user had asked relatives about family history beforehand.).

We have sought to clarify within the paper the methods and what was intended to be reported in this paper. There was a control clinic used for comparison of contemporaneous screening and referral patterns. The control clinic did not have access to MeTree and did not take part in any surveys on experience using MeTree reported in this paper.
3. It is not clear from this MS what data were concurrently collected from the control practice, how many subjects that practice enrolled, etc. Did that practice also use an exit questionnaire, for example? If so, how do the measured outcomes differ between this practice and the other two? If such a comparison was made, how did the analyses account for data-clustering at the practice level?

See above. The control clinic was used only to assess contemporaneous trends in screening and referrals. They did not enroll patients or participate in surveys and none of the data presented comes from the control clinic. We did calculate the design effect to control for clustering at the level of the clinic, which we report in the analysis section of this paper.

4. Flow diagram (Figure 1) implies that a large proportion of the subjects had not yet reached 12 months of follow-up. Why not delete the report of 12 month results, and wait until the data are complete? It seems to me that they add little that is new. The real “meat” of the 12-month follow-up ought to be the comparison of outcomes related to health and screening, between control and intervention participants, which are not reported in this paper.

We agree and have removed data on 12 month outcomes until more data is accrued.

5. “Ease of Use”: Readers interested in implementing MeTree or something like it would like to know how much time it took for the study staff to help people use MeTree, and how that varied by patient age. This is important for resource allocation.

We have added this data to the paper in paragraph 1 on page 9.

6. Outcomes related to patient reports of discussions with clinicians during a scheduled visit:

   a. Conclusions would be more interesting if a comparison were made between participants with and without MeTree. It seems that the investigators are precluding future publication of the effect of MeTree on patient-clinician discussions by reporting the data for only one group now. Perhaps the low response rate means that there is not adequate power for the planned comparison, in which case it might make sense to report this descriptive analysis here. If so, . .

   See response to question # 2. No data was obtained on provider discussions within control clinics.

   b. The article should clarify whether the low response rate for this “item” is, in fact, a low return rate for exit questionnaires, or something to do with this particular item on the survey.

   c. In either case, the low response rate limits the conclusions that can be drawn from the data. It is questionable whether to report them. But certainly, this limitation should be addressed front and center in the Discussion section.
d. Self-report, even in an exit questionnaire, of clinician-patient discussions is documented to be unreliable, when compared with direct observation of the encounter. This limitation should be acknowledged as well.

We have added clarification to the relevant paragraph in the discussion section for questions b-d.

7. **Statements in Discussion not substantiated by the data:** The authors and editors need to make sure that the Discussion and Conclusions represent the scientific findings of the investigators and others. Usually, unsubstantiated speculations could instead be phrased as 'important questions for further research, suggested by the findings from this study.' Examples: last sentence in second paragraph, page 13, regarding the likelihood that focusing on communication with family members would improve the family history obtained using MeTree.

The sentence was reworded to clarify the relationship of the data we collected to the statement.

8. **First sentence in third paragraph, page 13,** regarding MeTree improving providers’ discussions of risk-reducing recommendations. While it is possible, for example, that increasing preparatory communication with family members might help users’ FHH to be more accurate, it is also possible that this would have little effect, beyond the current method of implementation. That would be a question for future study.

We did state that further research was necessary; however, it may have been easily overlooked in the middle of the paragraph. We have added further clarification to the paragraph and emphasized the conclusion that further research was necessary by moving it to the end.

**Reviewer 2:**

**General comment (compulsory revision):**

1. **My major concern is that the authors do not cite any work supporting the evaluation of the public health use of this tool according to well-known criteria (Genetics in Medicine 2002; 4: 304-310):**
   - Analytical validity: how accurate and reliable is the information provided by a subject on the disease status of his/her relatives?
   - Clinical validity: does the familial risk stratification provided by the tool actually predict disease?
   - Clinical utility: can the knowledge of family health history promote the adoption of preventive behaviors?
   - Ethical/Legal/Social issues: do the benefits or revealing a positive family history for a disease outweigh the risks of having a label indicating risk for disease?

In my opinion, the authors should include a statement clearly affirming that this tool needs evaluation according to these ACCE (or similar) criteria before its use can be recommended for primary care settings. Meanwhile, the authors should be cautious in their recommendations.
We would like to note that the ACCE framework is meant to evaluate a diagnostic test- MeTree is not a diagnostic test, rather it is a program that allows implementation of evidence-based risk stratification within the primary care clinic environment; however, the question about analytic validity is a valid one and we have added references in the background to address the overall analytical validity of self-report FHH tools as compared to routine practice.

The clinical validity and utility of FHH has been extensively addressed in the evidence-based guidelines upon which MeTree is based. It was not the purpose of our study to review the guidelines (which have extensive documentation behind them) but to assist providers in implementing the guidelines. In terms of presenting the clinical validity and utility of MeTree, we are gathering data on this and describe this future step in the discussion section of the paper. The purpose of this paper is to address the acceptability and user experience of MeTree. Its impact on patient care hinges largely upon the ability to disseminate and sustain it, which is why we felt this topic warranted a manuscript.

Ethical/ legal/ social issues were added to the ACCE criteria in response to concerns about genomic/genetic diagnostic tests. While FHH is related to genetics, it is not embroiled in ELSI questions as it has been the standard of medical care for over a century, is recommended by every single medical body, and numerous studies have shown there is no anxiety related to sharing FHH data with providers. The data has been in the medical record and available to insurance companies, employers, etc… since the foundation of standardized medical practice. While it is now protected under GINA, it is standard of care and does not pose the concerns that newer diagnostic testing does.

Specific comments (minor essential revisions):

1. The authors claim to have based their risk stratification and recommendations on evidence-based published guidelines. However, several of the references they cite (13 to 19) do not seem to be guidelines and some of these references seem dated. On the other hand, guidelines are cited but not in this context (references 3 to 6).

We have updated the references to better reflect those actually used in the creation of the risk-stratification and clinical decision support within MeTree.

2. Even if the authors have based their risk algorithms and recommendations on published guidelines, it is unclear that these guidelines have based their risk assessment and recommendations solely on family health history. Is that the case?

MeTree collects all necessary information (FHH and personal history) to provide risk stratification and clinical decision support. We have added clarification to the first paragraph in the methods section.

3. The authors designed a clinic as control but data from this clinic do not appear anywhere in the manuscript (text, tables, figure).

See comments to reviewer 1 above. We have clarified the role of the control clinic in this study.
4. A large part of the results, pages 8-12, includes percentages but the authors reveal only the numerator of these percentages. Please add the denominators for the benefit of the reader.

We have added this detail to the results section.

5. The results include a series of odds ratios but in many instances it is not clear what groups are being compared. Moreover, some of the groups are labeled older, larger pedigrees, etc. without defining them.

We have clarified this within the analysis section of the paper.

6. In the sub-section “Statistical Analysis” (page 7) the authors assert that all calculations were assessed at a significance level of \( p < 0.05 \). It should say all of the hypotheses tests were performed at that level of significance.

This is already stated in the first sentence of the statistical analysis in the methods section.

7. Table 3 should include what the authors mean by routine and non-routine recommendations and provide examples of each. It will help if they add references to this classification.

Clarification was added to the statistical analysis paragraph in the methods section.

Thank you for the opportunity to submit our work to your journal. We appreciate your interest and feedback.

Sincerely,

R. Ryanne Wu