Author’s response to reviews

Title: Development of ReproKnow, a Reproductive Knowledge Assessment for Women with Rheumatic Diseases

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Author’s response to reviews:

Dear Reviewers and Editors,

Thank you most sincerely for reading our manuscript.

We have made changes based on the important comments and thoughtful feedback provided by the Reviewers, as follows:

Reviewer 1: Thank you very much for your positive review. We truly appreciate your commentary.

Reviewer 2: Thank you very much for your comments, which we address below:

1. In terms of the rationale and justification for the study - not enough case is made in your paper. You should justify the choice of the instrument. I don’t at all disagree with the study, but
the paper doesn't convince me as to why the ReproKnow was used, and that case needs to be made much more. In short, please explain why you have chosen to develop and validate the ReproKnow.

Thank you for this comment. Because many studies suggest that women with rheumatic diseases rarely receive any family planning counseling from providers, and other studies describe that few other relevant educational resources exist for these women, we wondered what women might know (or not know) about their reproductive health. Knowledge gaps could potentially translate to poor reproductive outcomes, especially as some adverse pregnancy outcomes may be influenced by patient behaviors (e.g., conceiving a pregnancy while using a fetotoxic anti-rheumatic drug). We posit that women cannot make well-informed reproductive decisions if they do not have some baseline reproductive knowledge. We therefore created ReproKnow as a tool that might help to clarify what these women know (and don’t know) about their diseases, anti-rheumatic drugs, and key reproductive health issues. The authors and a group of content experts identified the major knowledge domains that we felt might be particularly important for women with rheumatic diseases to know about their reproductive health.

As also mentioned in the Introduction, we hope to create educational interventions that will enhance women’s knowledge about their reproductive health; ReproKnow may help us to test the efficacy of our interventions. In the clinical setting, we also think ReproKnow may help rheumatologists or other health care providers to initiate a conversation about family planning and counseling with patients. By evaluating patients’ responses, health care providers may be able to identify important or relevant knowledge gaps, and educate their patients accordingly. Alternatively, patients may be activated to seek additional clarification about questions that were elicited from their use of ReproKnow.

These considerations informed our creation of a short-form instrument that could easily be used in research or in a clinical setting to capture what patients know about rheumatic diseases and reproductive health.

2. You should acknowledge if other instruments exist.

Thank you. The only other instruments that we are aware of were described in the original Discussion section [pg 13, line 282] as follows:

“Several reproductive knowledge assessments exist for specific diseases (e.g., Pregnancy in Rheumatoid Arthritis Questionnaire (PIRAQ)[26], and Crohn’s and Colitis Pregnancy Knowledge Score (CCP-Know)) [27]. However, such tools are not available for the majority of rheumatic diseases, including diseases with high pregnancy-associated mortality and morbidity, such as SLE and APS.”
Thus, while several other instruments to assess reproductive health knowledge are available for several specific autoimmune diseases, such instruments do not exist for a wide range of rheumatic conditions. This highlights another reason why ReproKnow may be helpful to rheumatologists and other health care providers, and researchers.

3. What the author(s) have done is different from the aims of the study. Objectives of the study sounded like or were phrased like activities (what was done, e.g., “describe the reproductive knowledge of a cohort of rheumatic disease patients”).

Thank you for your comment; we agree that our aims and objectives require further clarification. Our objectives were activity-based: 1) to develop and complete preliminary validation steps for a self-administered instrument (ReproKnow) to assess patients’ knowledge of pregnancy-related issues in the rheumatic diseases, 2) evaluate the use of ReproKnow in a community-based cohort of women of reproductive age with rheumatic diseases to test the hypothesis that many women with rheumatic diseases have suboptimal or poor knowledge of reproductive-related issues.

We have added this statement to the Introduction section [page 6, line 123], which now reads:

“To develop tailored interventions that ameliorate important gaps in patients’ reproductive knowledge, those gaps in knowledge must first be identified. Our objectives were to: 1) develop and complete preliminary validation steps for a self-administered instrument (ReproKnow) to assess patients’ knowledge of pregnancy-related issues in the rheumatic diseases; 2) evaluate the use of ReproKnow in a community-based cohort of women of reproductive age with rheumatic diseases, and describe patients’ reproductive knowledge using the tool.”

4. The Cronbach’s a values of the scale was low (below 0.70). I am aware that 0.7 is just a conventional cut-off, however, as it was stated “a coefficient of 0.7 or higher is generally considered to be acceptable for established scales.” Indeed, according to Peterson (1994, p. 381) “a scale in the preliminary stages of development is generally not thought to require the reliability of one used to discriminate between groups or of one being used to make decisions about individuals.” As the author(s) claim that the instrument is valid and ready to be used for educational interventions, this work could not be considered a preliminary development of a measure to assess the reproductive health knowledge of women with rheumatic diseases. Therefore, it is important to understand why Cronbach’s a was low. One possible reason is that the scale is not unidimensional but multidimensional. To this end, exploratory factor analysis (followed by confirmatory factor analysis) would be helpful. More important, Cronbach (1951)
indicated that, in presence of more than one dimension, the formula should be applied separately to items relating to different dimensions. In other words, first of all, the dimensionality of the scale should be examined. Cronbach’s alpha can be used for interval level variables. ReproKnow is not. All questions have 1 best answer except #10, which may have more than 1 best answer in addition to “Not sure”, it is not clear me if and how the answers were recoded. For instance, “not sure” was recoded as an incorrectly answered question?

Thank you very much for this comprehensive critique. First, we apologize if we inadvertently implied that ReproKnow is fully validated and ready to use for educational interventions. In the Abstract Background, we wrote that our objective was to “develop and validate” an assessment tool, which may be confusing. The original title also was, "Development and Validation of ReproKnow…", which also might be confusing.

What we actually did was to provide preliminary validation for a novel assessment test that we wish to use in a group of women with very high risk of adverse pregnancy outcomes. Our language through the original text better reflects the latter objective, including, “[Abstract] Initial testing of ReproKnow suggests that it may be a valid tool…”; “[Discussion] Our study provides initial evidence that ReproKnow is a valid tool for assessing reproductive knowledge…”; “ReproKnow is a tool that may help to evaluate the reproductive knowledge…”

We changed the title to, "Development and Initial Validation of ReproKnow". Since the Abstract Background sentence was also confusing, we have amended this section to read [page 3, line 48]:

“Our objective was to develop and preliminarily validate…”

To respond to the reviewer’s question about how we categorized answers, “Not sure” was recoded as an incorrectly answered question. We included this option to reduce the chances that a patient who wasn’t sure how to correctly answer a question selected an answer at random.

We agree with the reviewer that there are issues with the use of Cronbach alpha in our analysis. We also agree with the reviewer that use of Cronbach alpha may not be appropriate in this case because these are nominal-level items, and the alpha level is likely to underestimate the associations among variables. It is also unclear from a theoretical standpoint that knowledge is a unidimensional concept. Furthermore, the sample size is smaller than ideal for dimensional analysis (the standard would be a sample size of 300 patients).
To test these principles, we did complete the reviewer’s suggested analysis. First, we did a principal components analysis for nominal level variables and found four items on two dimensions that had no clinically meaningful or contextual relationships. Next, we removed these four items from the analysis altogether, and then removed all possible combinations of the items from the analysis. This did not change the alpha level—likely because of the nominal nature of the variables. The alpha for the four-item dimension was extremely low.

Given the results of the analysis suggested by the reviewer, we believe that Cronbach’s alpha is not an ideal test for this particular instrument. The factor analysis also was not helpful in identifying dimensions that are clinically interpretable. Conceptually, we believe this makes sense, as the topics covered by the questionnaire span many different topics across reproductive health. We therefore have removed the Cronbach’s alpha analysis from the Methods and the Discussion.

5. More information is also need on the sample (e.g., response rate) and the potential selective non-response (e.g., do survey respondents differ in their socio-demographic profile from those who declined or from what we know about women in western Pennsylvania?).

Thank you for this comment. We were not able to gather information about non-responders, which we did mention this as a limitation in the original Discussion section.

We appreciate the suggestion to give more information about the socio-demographic profile of western Pennsylvania. In our area, 82% are White, and adults with relatively low educational attainment (some college or less) comprise 50.3% of the sample.

Our sample was predominately White (77%) and had a similar level of education as the general western Pennsylvania population (some college or less: 49.7%). We incorporated these population descriptions into the Discussion [page 16, lines 339-343] as follows:

"First, while educational attainment and proportion of white participants in our sample were similar to the demographics of the general Western Pennsylvania population, the generalizability of our findings to women from other racial/ethnic or socioeconomic backgrounds may be limited."

6. If some women may have answered questions based on their own experiences, can the tool be revised to address this issue?

Thank you for this important comment. It is true that women’s responses may reflect their first-hand experiences, secondhand experiences (e.g., blog postings), prior family planning conversations with health care providers, etc. We cannot necessarily account for all of the
experiences that a woman has had that might have affected her knowledge about a specific domain tested in ReproKnow. In fact, if a woman had an atypical experience that led her to answer a question incorrectly (for example, if she became pregnant while using a highly effective contraceptive method, such as an intrauterine device, and subsequently downgraded the efficacy of that method when answering the contraception efficacy question in ReproKnow), our perspective is that there are no adverse consequences of a “wrong” answer on the assessment. We added the following comment to the Discussion section [page 16, lines 352-353]:

"Our perspective is that a “wrong answer” might actually provide an opportunity for a provider to clarify patients’ myths or misconceptions."

7. There is no evidence of criterion and content validity. This is not even mentioned in the Discussion section. The generalizability, or external validity, of the findings is not discussed. The findings of the study have been overstated. More research is needed to further validate and refine the tool.

Thank you for sharing this important comment. Criterion validity is the extent to which a measure is related to an outcome. While ReproKnow is not linked to any particular health outcome (and was not designed to necessarily be linked to outcomes), we could hypothesize that women who have more reproductive knowledge (and therefore may make well-informed health decisions) will have better pregnancy outcomes. This certainly could be studied in the future, perhaps in a longitudinal cohort of women for whom pregnancy outcomes could be tracked over time. Our cross-sectional design prevents us from being able to address criterion validity in the current analysis.

Regarding content validity, we formed a list of questions in addition to domains which we felt the questions addressed (as described in the paper, these included fertility, pregnancy outcomes, birth outcomes, safety of lactation, contraception, medication safety during pregnancy). As described in the original manuscript, “a group of local and national rheumatologists, obstetrician-gynecologists, internists with formal women’s health sub-specialization, nurses, a pharmacist, and a survey methodologist, reviewed each individual question.” We extracted 6 questions and refined the remaining questions.

We agree that additional validation steps could be used in the future to further ReproKnow’s validation. We took the reviewer’s suggestions and expanded our conversation about criterion and content validity, and the external validity of the findings, as follows [Discussion, page 16, 354-362]:

“In addition, more research is needed to further develop the psychometric properties of ReproKnow. Additional testing of the tool in a variety of clinical (e.g., community-based, academic, or hospital settings, and different geographic locations) or
research settings will help to further support the validity and reliability of ReproKnow. Our sample was not racially diverse, and the tool should be explored in more diverse populations of women with rheumatic diseases, perhaps with a wider range of rheumatic diseases. Criterion validity could be explored by assessing whether high scores on ReproKnow translate to better reproductive outcomes over time in a longitudinal cohort of women with rheumatic diseases.”

We truly appreciate the comprehensive, insightful, and thorough comments from our Reviewers. We hope that we have addressed all questions fully and are happy to answer any additional questions if they arise. Thank you most sincerely for your consideration.

The Authors