Author’s response to reviews

Title: Patient Perspectives on the Pathway to Psoriatic Arthritis Diagnosis: Results From a Web-Based Survey of Patients in the United States

Authors:

Alexis Ogdie (alogdie@pennmedicine.upenn.edu)
W. Benjamin Nowell (bnowell@ghlf.org)
Eddie Applegate (eddieapplegate@hotmail.com)
Kelly Gavigan (kgavigan@ghlf.org)
Shilpa Venkatachalam (svenky@ghlf.org)
Marie de la Cruz (marie.delacruz@iconplc.com)
Emuella Flood (emuellaflood@gmail.com)
Ethan J. Schwartz (ethan.schwartz@iconplc.com)
Beverly Romero (beverly.romero@iconplc.com)
Peter Hur (peter.hur@novartis.com)

Version: 2 Date: 18 Sep 2019

Author’s response to reviews:

September 12, 2019

Journal: BMC Rheumatology
Manuscript ID: BRHM-D-19-00033R1

Title: Patient Perspectives on the Pathway to Psoriatic Arthritis Diagnosis: Results From a Web-Based Survey of Patients in the United States

Dear Dr. Studenic,

I would like to thank the editorial office and reviewers for taking the time to provide thorough feedback on our submission. We have revised our manuscript in response to the reviewers’ comments below and are also submitting the updated manuscript for your consideration. Please note, any page or line numbers listed below correspond to the locations of each revision in the tracked changes version of the manuscript, which we have also attached for the editors.
We feel that the reviewers’ suggestions have been sufficiently addressed and hope that you will now consider this revised submission as suitable for publication in BMC Rheumatology.

Thank you again, and we look forward to hearing from you.

Kind regards,
Alexis Ogdie, MD, MSCE

Comments for the Author

Reviewer 1:

1. Introduction: I would welcome it if, in addition to the estimated prevalence, the total number of people affected were included too.

   Response: We thank the reviewer for this suggestion. We have revised the language on incidence and prevalence in the Introduction section – “Psoriatic arthritis (PsA) is a chronic inflammatory disease of the skin and musculoskeletal system with an estimated incidence of 6 per 100,000 and a prevalence of approximately 1 to 2 per 1000 in the general population” (Lines 57-58).

2. Methods: The authors write in the first paragraph: "Phase 1 consisted of targeted literature review and qualitative interviews with clinical experts and adults diagnosed with PsA via telephone to identify key concepts associated with disease burden and treatment experience. The key concepts identified were used to develop an online survey fielded to adults with PsA in Phase 2." I have some comments/questions to according this paragraph:
   a. I did not find a description of the literature review. Can you please provide more information? Keywords, databases, data extracted, ... ? Which data/questionnaires were used to create the questionnaire used in this study? Maybe as an appendix, if you have not enough words to do this in the Methods section.

   Response: Our targeted literature review consisted of a search in Embase and Medline. We have provided details of the search including the keywords, eligibility criteria, the literature review summary table, and the final questionnaire as a supplemental file for review only.

   b. Interviews with clinical experts/adults with PsA: Can you please provide more information? I did find the guideline in the appendix (Thank you for that) - but did not find anything about analysis, concepts, etc.

   Response: For clinical experts, interviews were transcribed, and a summary table of individual interview responses were developed to allow for comparisons of responses across experts. For
concept elicitation, the transcripts were analyzed using qualitative analysis software (MaxQDA, v.11). A codebook was developed based on the interview guide and responses were coded under relevant concepts (diagnosis, impacts, treatment burden, access to care, etc). We have provided summaries of the interview responses and concept elicitation as a supplemental file for review only.

c. Questionnaire: How was the questionnaire created in the end? How was the questionnaire tested? How did you test the validity and reliability of your questionnaire? Can you also please present the whole questionnaire as a supplemental file?

Response: Based on the findings from the targeted literature review, interview data, and concept elicitation interview data, we developed draft questions and response options, which included key concepts from the interviews with clinical experts and patients as well as two patient-reported outcome measures assessing current disease activity and quality of life (RAPID3 and PROMIS). An initial pilot test was then conducted with cognitive debriefing (CD) interviews to ensure clarity, comprehensiveness, and usability of the online surveys. The draft survey was tested using n = 3 patients per disease subgroup. These participants were interviewed about the content and any potential issues with the survey. The feedback from this first round of CD interviews were reviewed and changes were made upon agreement of the study team. Upon implementing the changes, an additional round of CD interviews was conducted with n = 2 patients per disease group.

d. Please check the reporting guidelines for conducting and reporting of survey research (such as the "Improving the quality of Web surveys: the Checklist for Reporting Results of Internet E-Surveys (CHERRIES)" or the guideline "Good practice in the conduct and reporting of survey research") and give all the necessary information. I would also appreciate it if you put the checklist in the appendix to enable the reader to quickly check the quality of your paper.

Response: The completed CHERRIES checklist has been uploaded for review.

e. You write: "All participants were required to provide verbal and electronic consent and authorization prior to participating." How did you obtain the oral consent of all 203 participants?

Response: We have revised the manuscript to clarify the consent process for our 2-phase study (Lines 87-92). In Phase 1, qualitative interviews were conducted with clinical experts and patients with PsA via telephone to identify key concepts associated with disease burden and treatment experience; the information obtained was used to develop an online survey fielded to adults with PsA in Phase 2. We obtained verbal consent from interviewees in Phase 1, and electronic consent from survey respondents in Phase 2 as outlined in our IRB protocols which were approved by a central review board, Salus IRB. Both IRB approvals were uploaded as supplementary materials for review.
f. You write: "Continuous variables were presented using means and SDs" - were these data normally distributed? If not please provide median and range too.

Response: We have added the following language to our manuscript (Lines 167-168): “Due to the large sample size and due to the central limit theorem, the sample means were assumed to be normally distributed.”

g. Did you correct for multiple testing? If yes, please describe it (Bonferroni? Bonferroni-Holm?). If not - please give a statement of why you did not. Because you did a lot of testing.

Response: We have added the following language to our manuscript (Lines 168-170): “No Bonferroni or other correction for multiple testing was performed due to the preliminary/exploratory nature of these analyses, which may form the basis of hypotheses in future studies.”

3. Results:
a. Please be more clear and display also values in the text (especially the p-values, because they are also missing in the tables), especially if you talk of "differences".

Response: We thank the reviewer for this suggestion. We have revised the text (Lines 192-195, 209-212) and both tables to include the P values for clarity.

b. Please provide p-values in the tables, especially if you did no correction for multiple testing.

Response: Consistent with the changes to the text noted above, we have revised both tables to include the P values for clarity.

c. You had 203 respondents - what percentage is that to the total number of people affected?
Response: We have provided more information on the recruitment process (Lines 140-144). Of a total of 572 unique email views, the number of respondents who clicked to view the first page of the survey was 326, resulting in a view rate of 57.0%. Of the 326 visitors to the survey site, 258 unique visitors agreed to participate and initiated the survey, resulting in a participation rate of 79.1%. A total of 203 respondents completed the questionnaire for a completion rate of 78.7%.

Reviewer 2:

1. Both in the abstract and in the beginning of the methods you refer this was a "Two phase study". The reason for this is completely understandable and correct. My questions are:
   a. Have you considered to designate this as a "Mixed-methods study"? I would suggest you to read this editorial from Elisabeth Halcomb - Mixed methods research: The issues beyond combining methods - Journal of Advance Nursing. https://doi.org/10.1111/jan.13877
Response: We thank the reviewer for this suggestion. We have revised Line 87 and introduced our study as a “mixed-method, 2-phase study.”

b. You only report the results of the quantitative study. Have you considered to present, even briefly, the qualitative part? I would be in favor of it, even if mainly in Supplementary material. Are you considering to present those results in another publication?

Response: We thank the reviewer for noting this. At present, we would like to focus on Phase 2 of the analysis, which provides more pertinent information applicable to the overall care and management of patients with PsA. However, all materials pertaining to the execution of Phase 1 have been provided as supplemental files for editorial review only.

2. Also in the methods section, in the end of the description of "study variables" you correctly described the stratification of time to diagnosis into 3 categories. In the following section ("Data analysis") you seem to repeat part of this information: "Data were additionally stratified by time to PsA diagnosis using χ² tests to analyze differences across the groups". This sentence seems a bit confusing. Maybe instead of "using chi-square" you may say "and chi-square was used to".

Response: We thank the reviewer for this suggestion. We have revised this sentence for clarity (Lines 165-167).

3. You report a total of 203 respondents. Which was the response rate?
Response: We have provided more information on the recruitment process (Lines 140-144). Of a total of 572 unique email views, the number of respondents who clicked to view the first page of the survey was 326, resulting in a view rate of 57.0%. Of the 326 visitors to the survey site, 258 unique visitors agreed to participate and initiated the survey, resulting in a participation rate of 79.1%. A total of 203 respondents completed the questionnaire for a completion rate of 78.7%.

4. Which was the period of data collection? This is important to put the conclusion in (time) context.

Response: We have provided more information on the recruitment process (Lines 144-145). Emails were first sent on September 5, 2017, and the final response was received on October 13, 2017, totaling 38 days of data collection.

5. In the results section, page 8, last sentence of first paragraph, reporting the comparison of characteristics between time to diagnosis group, I did not make the same interpretation of the Table 1 results. For instance, the % of respondents with high school education was: 4.3% (< 6 months), 1.5% (6m to 4y), and 9.1% (≥5y of diagnosis). but you reported that faster times to diagnosis (< 6m) appeared to ... have higher education...

Response: We thank the reviewer for noting this. We have revised the sentence in the manuscript to read “Respondents with faster times to diagnosis (< 6 months) appeared to be slightly older,
have undergraduate/postgraduate education, were less likely to be disabled and not working, and were more likely to earn ≥ $100,000/year compared with respondents with longer times to diagnosis” (Lines 181-182).

6. Regarding Table 1, the "italics" to highlight statistically significant differences is hard to be seen/differentiated.

Response: We thank the reviewer for noting this. We have revised both tables to include the P values for clarity.