

PEER REVIEW HISTORY

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ARTICLE DETAILS

TITLE (PROVISIONAL)	Factors Associated with Patient-reported Likelihood of Using Online Self-care Interventions: A Scleroderma Patient-centered Intervention Network (SPIN) Cohort Study
AUTHORS	Kwakkenbos, Linda; Cumin, Julie; Carrier, Marie-Eve; Bartlett, Susan; Malcarne, Vanessa; Mouthon, Luc; Nielson, Warren; Rannou, F; Welling, Joep; Thombs, Brett

VERSION 1 – REVIEW

REVIEWER	Elizabeth Wahl University of Washington VA Puget Sound Healthcare System USA
REVIEW RETURNED	28-Feb-2019

GENERAL COMMENTS	<p>The authors are to be congratulated for their extensive work on scleroderma. This study evaluates the association between scleroderma patients' demographic, disease and clinical features and their self-reported willingness to participate in online self-management programs in a large multi-national cohort (SPIN). The rationale for the study is fairly clear: scleroderma is a rare disease, and in order to optimize efficiency of studying patients, novel designs such as the cohort multiple RCT (cmRCT) are now being used. One potential limitation of the cmRCT approach is challenges with patients agreeing to participate in studies not initially described to them; the authors thus seek to understand factors influencing patients' self-reported willingness to enroll in research directly applicable to their condition. Overall, this is an important question. The authors' primary finding is that patients' overall likelihood of using an online intervention (rather than demographic, clinical, or disease-specific factors) is the strongest predictor of self-reported willingness to participate in online self-management. Importantly this suggests that perhaps a more efficient (and less burdensome) questionnaire may be used to assess participants' willingness to participate in research. However, this study does not evaluate the relationship between self-reported willingness to participate and actual participation. Overall, it would benefit from some editing for clarity. A few more specific comments:</p> <ol style="list-style-type: none">1. While the abstract introduces the idea of signaling questions, it is a bit confusing that there is no explicit mention of measures in the methods section of the abstract. It may be useful to readers and improve the clarity of the abstract to make the objectives section more concise and include a measure section specifying the data collected (demographic, clinical, PRO, and patient self-reported willingness to participate in online self-
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	<p>management programs). It may also be useful to clarify how general 'willingness to participate' was assessed.</p> <p>2. In the main body of the paper, Methods/Statistical Analysis section, on page 17/line 43 – would include a description of how correlations reported in Table 2 were assessed.</p> <p>3. Page 17/line 43 – I think it would be clearer when you are describing the multi-regression analysis to be clearer with section (b) on line 54 - for example: "To assess factors associated with self-reported likelihood of participating in an online program, we conducted multiple linear regression analysis for each signaling question in a stepwise fashion. Independent variables included in the regression models were determined apriori, and included (a)...(b) general likelihood of using online interventions, calculated by taking the mean score of the remaining signaling questions, (c)..." etc.</p> <p>4. Page 17/line 43, I think you mean 'likelihood' not 'likeliness'</p> <p>5. Results section – I notice that you never report mean scores for SEMCD, PROMIS-29v2 domains, CHFS, SAAS, GI interference, or physical activity scales. Consider including this in table 1, or in the appendix.</p> <p>6. Signaling question scores all cluster around the middle, skewed towards willingness to participate (5-7, most in the 6 range). This likely impacted your findings, may be worth mentioning. Were you able to do a sensitivity analysis, exploring characteristics of those least (or most) willing to participate?</p>
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REVIEWER	E Jury University College London, UK
REVIEW RETURNED	09-Aug-2019

GENERAL COMMENTS	This is an interesting study assessing factors influencing patient participation in online interventions. This is a very topical and novel approach and I have no specific comments about the study design of results.
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VERSION 1 – AUTHOR RESPONSE

Reviewer #1:

The authors are to be congratulated for their extensive work on scleroderma. This study evaluates the association between scleroderma patients' demographic, disease and clinical features and their self-reported willingness to participate in online self-management programs in a large multi-national cohort (SPIN). The rationale for the study is fairly clear: scleroderma is a rare disease, and in order to optimize efficiency of studying patients, novel designs such as the cohort multiple RCT (cmRCT) are now being used. One potential limitation of the cmRCT approach is challenges with patients agreeing to participate in studies not initially described to them; the authors thus seek to understand factors influencing patients' self-reported willingness to enroll in research directly applicable to their condition. Overall, this is an important question. The authors' primary finding is that patients' overall likelihood of using an online intervention (rather than demographic, clinical, or disease-specific factors) is the strongest predictor of self-reported willingness to participate in online self-management. Importantly this suggests that perhaps a more efficient (and less burdensome) questionnaire may be used to assess participants' willingness to participate in research. However, this study does not evaluate the relationship between self-reported willingness to participate and actual participation. Overall, it would benefit from some editing for clarity. A few more specific comments:

We thank Reviewer #1 for her enthusiasm for the study and the helpful comments that follow.

1. While the abstract introduces the idea of signaling questions, it is a bit confusing that there is no explicit mention of measures in the methods section of the abstract. It may be useful to readers and improve the clarity of the abstract to make the objectives section more concise and include a measure section specifying the data collected (demographic, clinical, PRO, and patient self-reported willingness to participate in online self-management programs). It may also be useful to clarify how general 'willingness to participate' was assessed.

We have done our best to address this comment by adding more details on the measures, but were constrained by the journal's permitted maximum word count for the abstract (no more than 300 words). We have added the following information to the abstract (Page 9, Lines 15-17):

"Measures: Demographic and disease characteristics, self-efficacy, and symptoms related to each specific intervention were completed in addition to signalling items. General likelihood of using interventions was calculating by taking the mean score of the remaining signalling questions."

2. In the main body of the paper, Methods/Statistical Analysis section, on page 17/line 43 – would include a description of how correlations reported in Table 2 were assessed.

We have added the following information to the Statistical Analysis section of the manuscript (Page 17, Line 18):

"Pearson correlations between signalling question scores were calculated."

3. Page 17/line 43 – I think it would be clearer when you are describing the multi-regression analysis to be clearer with section (b) on line 54 - for example: "To assess factors associated with self-reported likelihood of participating in an online program, we conducted multiple linear regression analysis for each signaling question in a stepwise fashion. Independent variables included in the regression models were determined apriori, and included (a)...(b) general likelihood of using online interventions, calculated by taking the mean score of the remaining signaling questions, (c)..." etc.

We thank reviewer #1 for this suggestion and have edited the text of the manuscript accordingly, except for using the term "hierarchically" instead of "stepwise" to avoid confusion with stepwise regression methods (i.e., that there were data-driven steps for inserting variables). The text now reads (Page 17-18, Lines 18-1):

"To assess factors associated with self-reported likelihood of participating in an online program, we conducted multiple linear regression analysis for each signalling question and entered sets of variables hierarchically. Independent variables included in the regression models were determined a-priori, and included: (a) demographic and disease characteristics including age, sex, disease duration (time since onset of first non-Raynaud symptom), modified Rodnan Skin Score, years of education; (b) general likelihood of using online interventions, calculating by taking the mean score of the remaining signalling questions;..."

4. Page 17/line 43, I think you mean 'likelihood' not 'likeliness'

We thank the reviewer for pointing this out and have corrected the text accordingly.

5. Results section – I notice that you never report mean scores for SEMCD, PROMIS-29v2 domains, CHFS, SAAS, GI interference, or physical activity scales. Consider including this in table 1, or in the appendix.

We have added the means (SD) for the PROs to Table 1, as per the reviewer's suggestion.

6. Signaling question scores all cluster around the middle, skewed towards willingness to participate (5-7, most in the 6 range). This likely impacted your findings, may be worth mentioning. Were you able to do a sensitivity analysis, exploring characteristics of those least (or most) willing to participate?

We thank reviewer #1 for this suggestion. To address this comment, we have added the dispersion with the response frequencies to the Appendix of the paper and commented on the distribution of scores in the manuscript (Page 20, Lines 6-8):

“Response frequencies for signalling items are shown in Appendix Table A. Responses for each signaling question were skewed towards willingness to participate, with score 10 (very likely to participate) being most frequently given for all 9 items (range 22-36%).”

Reviewer #2

This is an interesting study assessing factors influencing patient participation in online interventions. This is a very topical and novel approach and I have no specific comments about the study design of results.

We thank Reviewer #2 for her/his enthusiasm for the study.