

PEER REVIEW HISTORY

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ARTICLE DETAILS

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| TITLE (PROVISIONAL) | An international multiphase mixed methods study protocol to develop a patient-reported outcome instrument for children and adolescents with lower limb deformities |
| AUTHORS | Chhina, Harpreet; Klassen, Anne; Kopec, Jacek; Oliffe, John; Cooper, Anthony |

VERSION 1 - REVIEW

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| REVIEWER | Antoni Fellas PhD Candidate - B HSc (Hons) B Pod The University of Newcastle, School of Health Sciences, Faculty of Health and Medicine - Callaghan, NSW, 2308, Australia. Email: antoni.fellas@uon.edu.au |
| REVIEW RETURNED | 12-Nov-2018 |

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| GENERAL COMMENTS | <p>Firstly I would like to congratulate the authors on an well written and structured paper.</p> <p>Overall I would recommend this paper for publication following some minor revisions to the submitted manuscript:</p> <p>Introduction: I have the following queries with the introduction section:</p> <ol style="list-style-type: none">1. To provide some context to the paper and the significance of developing a tool for lower limb deformities, I believe it would be prudent to include: a definition of lower limb deformities (or what that may include); the implications of these deformities (physical/psychological/social/financial); prevalence rates (if known). I know providing detailed information on lower limb deformities is not the aim of this paper but providing a few basic sentences on lower limb deformities may capture the impact of deformities have on children and adolescents (to the reader), and why it is important to develop a valid and reliable tool.2. I would also recommend being more explicit in the rationale/justification section of the introduction. Specifically, why do we need this tool? (measuring impact of interventions?) and how it could be beneficial for health professionals? What type of health professionals may this tool be targeted for? e.g. orthopaedic surgeons/specialists, allied health professionals |
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| | <p>(physio/nursing/OTs/podiatry) or both? This was somewhat touched on in the last section of the paper but I believe this may require more attention and made more explicit in the introduction section, and not just at the end of the paper.</p> <p>Methods: The methodology has been well presented and adequately supported by references. Well done. Only have the following queries:</p> <ol style="list-style-type: none"> 1. Given the tool will be specific to the lower limb, kindly wondering if the authors considered having clinical input from a podiatrist regarding feedback on the lower limb scales? 2. Recruitment/Participants section: Just wanted to clarify the diagnosis of lower limb deformities and who they are made by? I'm assuming all diagnoses are made clinically by orthopaedic surgeons in the respective countries? If so, do they have a standardised criteria (if one exists) to diagnose these problems or are they all made purely by their clinical discretion? It may be helpful to the reader to know that the participants were diagnosed accurately/gold standard way. <p>Spelling Mistake(s) Under Phase 3 Ethics 3rd line - Maintaining and Challenges spelled incorrectly (Page 18, line 22)</p> <p>References appear to be in correct order.</p> <p>Thank you kindly for the opportunity to review this manuscript.</p> |
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| REVIEWER | Walter Ricciardi Università Cattolica del Sacro Cuore, Institute of Public Health, Rome, Italy |
| REVIEW RETURNED | 10-Jan-2019 |

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| GENERAL COMMENTS | <p>This research protocol addresses a very important issue regarding the development of a patient-reported outcome (PRO) instrument for children and adolescents affected by lower limb deformities. The protocol describes three phases needed to develop a new PRO instrument and is generally clear and well written. I would suggest some minor revisions:</p> <p>Introduction In the introduction section the Authors highlight the lack of availability of this specific instrument in scientific literature. This is confirmed by a systematic review that they recently carried-out. This section should be completed by reporting epidemiologic data on lower limb deformities and their impact in terms of health-related quality of life in particular for children and adolescents. Adding this information will be useful to better define the research context.</p> <p>Methods Phase 1; Step1 1. The description of the qualitative interviews appears quite fragmented. I suggest that a separate paragraph to describe</p> |
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| | <p>qualitative interviews be created in order to avoid this fragmentation.</p> <p>2. Page 7 line 9. These two sentences are not very clear. It seems that one “lead interviewer” will conduct all the interviews in 3 centres (Canada, USA and India), two English speaking and one Indian Hindi/Punjabi speaking centre, at main study site. What the Authors mean with “main study site”? (e.g. one main study site per Country or a unique main study site; it should be specified what is the main study site) And, what is the native language of the lead interviewer?</p> <p>3. More details about the interview guide and about its translation process should be also included (e.g. Are all the translations performed by the same person? And, if yes, what is his native language?).</p> <p>4. Furthermore, the process of audio-recording, transcription and translation of the interviews also needs some clarification (e.g. who will perform the translation, and what is his native language). Phase 1; Step3</p> <p>5. Page 10 line 17. The translation process is well described, but it would be useful to specify the native language of researcher/s involved in this process.</p> <p>6. Page 13 line 1-2. The Authors report that the data collection (PRO instrument completion) in this phase will be based at the discretion of each site, paper or electronic version. Is the instrument self-administered by the patient in any case? Which are the data sources for demographic and clinical data? Phase 3</p> <p>Test-retest reliability: how will the Authors establish which participants present stable leg condition? Furthermore, the Authors will calculate Intraclass Correlation Coefficient: will the final score be a continuous score?</p> <p>Construct validity: Although the Authors added a reference, it could be useful to specify in the text which will be the HRQOL generic instrument that they intend to use.</p> <p>Eventually, there are some typos that need revision: Page 6 line 6. “item pool that is used to information a set of preliminary scales”. Do the Authors mean “item pool that is used to inform a set of preliminary scales”?</p> <p>Page 15 line11 “assessment!”</p> <p>Page 15 line16 “instrumentwill”</p> <p>Page 16 line 15. The Authors write “three groups” but then list four groups “no improvement, small, moderate and large improvement”. Is this a typo?</p> <p>Page 16 line 18. “A distribution based approach will be also be used”</p> <p>Page 18 line 4. “the effectiveness of interventions in terms improving the patient HRQOL”.</p> |
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VERSION 1 – AUTHOR RESPONSE

Reviewer(s)' Comments to Author:

Reviewer: 1

Reviewer Name: Antoni Fellas

Institution and Country: PhD Candidate - B HSc (Hons) B Pod

The University of Newcastle, School of Health Sciences, Faculty of Health and Medicine - Callaghan, NSW, 2308, Australia.

Email: antoni.fellas@uon.edu.au

Please state any competing interests or state 'None declared': None declared

Please leave your comments for the authors below

Firstly I would like to congratulate the authors on an well written and structured paper.

Overall I would recommend this paper for publication following some minor revisions to the submitted manuscript:

Introduction:

I have the following queries with the introduction section:

1. To provide some context to the paper and the significance of developing a tool for lower limb deformities, I believe it would be prudent to include: a definition of lower limb deformities (or what that may include); the implications of these deformities (physical/psychological/social/financial); prevalence rates (if known). I know providing detailed information on lower limb deformities is not the aim of this paper but providing a few basic sentences on lower limb deformities may capture the impact of deformities have on children and adolescents (to the reader), and why it is important to develop a valid and reliable tool.

Reply: Thank you for your suggestion. This information has now been added to the introduction section of the manuscript.

2. I would also recommend being more explicit in the rationale/justification section of the introduction. Specifically, why do we need this tool? (measuring impact of interventions?) and how it could be beneficial for health professionals? What type of health professionals may this tool be targeted for? e.g. orthopaedic surgeons/specialists, allied health professionals (physio/nursing/OTs/podiatry) or both? This was somewhat touched on in the last section of the paper but I believe this may require more attention and made more explicit in the introduction section, and not just at the end of the paper.

Reply: Thank you for your suggestion. This information has now been added to the introduction section of the manuscript.

Methods:

The methodology has been well presented and adequately supported by references. Well done. Only have the following queries:

1. Given the tool will be specific to the lower limb, kindly wondering if the authors considered having clinical input from a podiatrist regarding feedback on the lower limb scales?

Reply: Thank you for your valuable suggestion. We have now incorporated getting feedback from podiatrists along with the other healthcare professionals involved in the care of children with lower limb deformities.

2. Recruitment/Participants section: Just wanted to clarify the diagnosis of lower limb deformities and who they are made by? I'm assuming all diagnoses are made clinically by orthopaedic surgeons in the respective countries? If so, do they have a standardised criteria (if one exists) to diagnose these problems or are they all made purely by their clinical discretion? It may be helpful to the reader to know that the participants were diagnosed accurately/gold standard way.

Reply: All participants included in this study are/will be diagnosed by the treating orthopaedic surgeon in their respective countries. There is no standardised criterion for diagnosis of lower limb deformities. Each of this condition is diagnosed by a combination of clinical examination and radiological investigations.

This has been clarified in the section labelled as 'inclusion and exclusion criteria'.

Spelling Mistake(s)

Under Phase 3

Ethics

3rd line - Maintaining and Challenges spelled incorrectly (Page 18, line 22)

Reply: Thank you for bring this to our attention. These errors have now been fixed.

References appear to be in correct order.

Thank you kindly for the opportunity to review this manuscript.

Reviewer: 2

Reviewer Name: Walter Ricciardi

Institution and Country: Università Cattolica del Sacro Cuore, Institute of Public Health, Rome, Italy

Please state any competing interests or state 'None declared': None declared

Please leave your comments for the authors below

This research protocol addresses a very important issue regarding the development of a patient-reported outcome (PRO) instrument for children and adolescents affected by lower limb deformities. The protocol describes three phases needed to develop a new PRO instrument and is generally clear and well written. I would suggest some minor revisions:

Introduction

In the introduction section the Authors highlight the lack of availability of this specific instrument in scientific literature. This is confirmed by a systematic review that they recently carried-out. This section should be completed by reporting epidemiologic data on lower limb deformities and their impact in terms of health-related quality of life in particular for children and adolescents. Adding this information will be useful to better define the research context.

Reply: Thank you for your suggestion. This information has now been added to the introduction section of the manuscript.

Methods

Phase 1; Step1

1. The description of the qualitative interviews appears quite fragmented. I suggest that a separate paragraph to describe qualitative interviews be created in order to avoid this fragmentation.

Reply: Thank you for your valuable suggestion. We do agree that the qualitative information is fragmented. However, the qualitative components are incorporated in the manuscript according to their respective phases of the PRO instrument development not by research methodology.

2. Page 7 line 9. These two sentences are not very clear. It seems that one “lead interviewer” will conduct all the interviews in 3 centres (Canada, USA and India), two English speaking and one Indian Hindi/Punjabi speaking centre, at main study site. What the Authors mean with “main study site”? (e.g. one main study site per Country or a unique main study site; it should be specified what is the main study site) And, what is the native language of the lead interviewer?

Reply: The information requested has been added to the manuscript and is also presented below.

The main study site is the site where the first and the senior author are located.

The native language of the lead interviewer is Punjabi and Hindi. The lead interviewer is fluent in English.

3. More details about the interview guide and about its translation process should be also included (e.g. Are all the translations performed by the same person? And, if yes, what is his native language?).

Reply: More details about the interview guide and its translations have been added (page 10)

4. Furthermore, the process of audio-recording, transcription and translation of the interviews also needs some clarification (e.g. who will perform the translation, and what is his native language).

Phase 1; Step3

Reply: More information about the transcription and translation of interviews has now been added to the manuscript (page 11).

5. Page 10 line 17. The translation process is well described, but it would be useful to specify the native language of researcher/s involved in this process.

Reply: Details about the native language of the researchers involved have been added.

6. Page 13 line 1-2. The Authors report that the data collection (PRO instrument completion) in this phase will be based at the discretion of each site, paper or electronic version. Is the instrument self-administered by the patient in any case? Which are the data sources for demographic and clinical data?

Phase 3

Reply: The instrument is designed to be self-administered. However, not all of the participating sites will have the resources to have the data collected and entered directly into the RedCap database.

Along with the consent form, each site will also complete a demographic form for each participant being interviewed. The basic demographic and clinical data will be obtained from this demographic sheet.

Test-retest reliability: how will the Authors establish which participants present stable leg condition?

Reply: Confirmation of the stable leg condition will be obtained from the treating orthopaedic surgeon at each of the participating sites.

Furthermore, the Authors will calculate Intraclass Correlation Coefficient: will the final score be a continuous score?

Reply: Thanks for your question. Yes, the final score will be a continuous score.

Construct validity: Although the Authors added a reference, it could be useful to specify in the text which will be the HRQOL generic instrument that they intend to use.

Reply: This has been added as suggested.

Eventually, there are some typos that need revision:

Reply: Thanks for bringing these to our attention. These typos have now been corrected.

Page 6 line 6. "item pool that is used to information a set of preliminary scales". Do the Authors mean "item pool that is used to inform a set of preliminary scales"?

Page 15 line11 "assessmentl"

Page 15 line16 "instrumentwill"

Page 16 line 15. The Authors write "three groups" but then list four groups "no improvement, small, moderate and large improvement". Is this a typo?

This has been replaced by 'four groups' in the manuscript.

Page 16 line 18. "A distribution based approach will be also be used"

Page 18 line 4. "the effectiveness of interventions in terms improving the patient HRQOL".

VERSION 2 – REVIEW

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| REVIEWER | Antoni Fellas University of Newcastle, Australia |
| REVIEW RETURNED | 10-Feb-2019 |

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| GENERAL COMMENTS | Dear Authors, Thank you for revising your manuscript to address both sets of reviewer comments. I have read through your revised manuscript and have no further comments to add. I would therefore recommend this for publication. Congratulations on a well written paper. All the best with your study. |
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| REVIEWER | Walter Ricciardi Università Cattolica del Sacro Cuore, Sezione di Igiene, Istituto di Sanità Pubblica, Rome, Italy |
| REVIEW RETURNED | 12-Feb-2019 |

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| GENERAL COMMENTS | Thanks to the Authors for their accurate response. The revised manuscript addresses properly all my requests for clarification. I have no further comments. |
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