Author’s response to reviews

Title: Development of a core outcome set for orthodontic trials using a mixed-methods approach: protocol for a multicentre study

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Reviewer reports:

Reviewer #1: This is a very interesting study protocol that addresses an important limitation in RCT in dentistry. Poor reporting, lack of standardized outcome measures and the selection of inappropriate outcomes make it difficult any attempt to combine data in systematic reviews and metanalysis and limit the whole potential of well-conducted clinical studies to provide valuable information for patients, healthcare providers and health managers.

The relevance of this issue in Orthodontics was clearly summarized in the introduction, considering both the ethical and economic aspects of clinical research in health sciences.

Some minor comments that should be addressed by authors:

1 - The aim of the first stage (scoping review) should be clearly stated - this is mentioned only in the abstract. In addition, a brief description of the main characteristics of a scoping review (and
how it differs from a systematic review) would be useful for readers to understand that why it was chosen as the hypothesis-generating strategy for this study protocol.

Thank you for the comments. The aim and a brief description of the scoping review been incorporated in the main text:

‘Scoping reviews are used to map key concepts underpinning a broad research area and are useful for examining emerging evidence [19]. Although scoping reviews follow a systematic approach to evidence gathering, they differ from systematic reviews in that they aim to present a broad overview of evidence relating to a topic irrespective of study quality, rather than focusing on answering a specific question based on carefully selected study designs [20]. The aim of this scoping review is to identify outcomes employed in contemporary orthodontic research, which will partly be used to inform the long list of outcomes for the Delphi surveys.’

2 - Please give a more detailed information about the Study Advisory Group (definition, components, duties...).

This has now been highlighted in the manuscript:

‘The grouping of outcome measures and outcome domains will be reviewed by the Study Advisory Group (SAG), which will consist of four clinicians with a range of expertise in the fields of orthodontics and patient-centred research as well as two qualitative researchers and a patient representative. The SAG will oversee key stages and help shape conclusions of the project.’

3 - The rationale for adoption a mixed-methods approach should be considered in the discussion section, reinforcing the complimentary aspects of the different steps of a multimethod research. The use of a quantitatively driven approach may also be justified, considering its advantages to provide more complex answers to the research questions, and by using high quality qualitative data derived from well planned data collection and analysis.
Thank you for the suggestion. We had initially kept the Discussion to a minimum in keeping with typical Trials submissions but have now expanded upon this to include the rationale for and benefits of mixed-methods research as suggested:

‘In theory, a simpler method, such as a questionnaire, could be used to gather information from patients, as was the case in the COS development for childhood asthma, which had the advantage of minimising the burden on participants and enabling the involvement of a larger participant sample [30]. However, for the present research, it was felt that it would be best if unprompted patient views and attitudes concerning orthodontic outcomes were obtained, rather than providing quantified answers to preconceived ideas as would be the case with a clinician-derived questionnaire [31]. Additionally, questionnaires do not offer opportunities to clarify ambiguous data and equally there is no means of knowing if participants fully understood or even if they misunderstood questions posed. Finally, since this COS is directed at children and young people, it was also felt that their opinions would require in-depth analysis in order to be able to subsequently convert these opinions into outcomes. Consequently, it was felt that this research question would best be addressed using an integration of quantitative and qualitative data (i.e. mixed-methods). A similar approach was also used in determining key health outcomes and development of a COS for children and young people with neurodisability [32].’

Reviewer #2: This study aims to standardize outcome measures in orthodontic trials, it relies on a mixed methods approach. The manuscript is well written. I only have very few comments and I also need that authors clarify these following points:

- I suggest to remove the abbreviations from the abstract (NHS -line 9 and COMET -line 36) and replace them but the whole terms/sentences.

The abbreviated words have been replaced with the whole terms.

-In the background:

Line 25: The abbreviation "NHS" appears for the first time in the text, but has not been defined before. So replace by: National Health Service (NHS)
This has been amended as suggested.

In the second paragraph, the authors show the relevance of standardizing outcomes for effectiveness and intervention research (4th paragraph, which is very pertinent to highlight the need for their study. However, authors also mention the disadvantage of using the IOTN for defining eligible patients to NHS orthodontic treatment (2nd paragraph): An opinion that I approve completely, but I don't see how the results of this study will be useful in changing that issue? It looks like the study defines the orthodontic outcomes from patients and their parents' perspectives. However, the need of orthodontic treatment depends on the clinical evaluation, not on the patient perception only. I think that patient perception is specially important to know what patients expect from a treatment, so we can take their expectations into account as clinicians and researchers. I may be wrong, but I have a real doubt that NHS will rely on these kind of outcomes, rather than clinical criteria, to establish its eligibility criteria for orthodontic treatment.

Thank you for your comments. We agree that this is likely to be the case, however the purpose of highlighting the IOTN in the manuscript is to illustrate that patient expectations and expressed need are often different from that of the clinicians. We agree that this research is unlikely to change that but it may be instructive for future researchers and may help to introduce patient perceptions through a validated measure as part of the assessment/eligibility process in the future.

-Methods/Design:

For the first paragraph of this section (line 56-58), that is related to ethical approval: I suggest to move it to the end of the section. First, the reader has to have an idea about the methodology used in the study, to justify the ethical approvals that were needed/obtained.

Thank you. The ethical approval paragraph has been moved to end of the methodology section.

Stage 1: Scoping review: In the section Participants (Line 15), I suggest to specify either a range of age or at least the maximum age for the participants.
We have amended this accordingly.

Stage 3: Delphi surveys - Parents and young people sample (line 29): are these participants (parents and children) different from the ones who participate to stage 2? it's not clear here! Also, since this stage 3 will provide quantitative data for quantitative analysis, did you estimate a sample size for this stage 3 (for both groups: Clinicians and parents-young people?)

Thank you for the comments. Yes, the sample here will be different to the previously recruited sample. This has now been made clear in the text. A sample size has not been estimated for the Delphi surveys but the advice from COMET is to have a minimum of 10 participants per stakeholder group, with previous similar research having over 70 participants in total in all 3 rounds (Harman et al, 2015).

Also, authors will rely on the SAG in different steps of the study, is it possible to clarify who are the members of SAG (are they researchers and experts only, or also patients) ?

This has now been clarified in the text as suggested in line with both your comment and those of Reviewer 1.

The last paragraph of stage 3: Final data analysis: is the classification that will be used for the consensus based on some previous research? there is no reference.

Yes, it is based on previous research and the reference has now been added there. Thank you for the comment.

Stage 4: Consensus meeting: About the first sentence line 13 "Finally, consensus meetings will be held with all stakeholders involved (patients, parents,clinicians and commissioners)". All along the text before, I didn't see the commissioners being part of the study procedures. I understand that researchers may involve them when the study comes to this stage (4). But, even in this part,it's not explained how they will be part of the study. There are no details about which commissioners will be participating, and how they will do? In fact, the use of the term of "stakeholders" in the manuscript will be inappropriate and confusing if only parents, children and clinicians will participate to the study.
Depending on the authors' answer to these question, it may be necessary to review the table 1, because it's mentioned in the last line Consensus meeting (service users, providers and policy makers)

Thank you for the comments. We aim to include commissioners and clinicians actively involved in orthodontic service commissioning only in this stage of the research (stage 4). We plan to engage with National Health Service dental commissioners during the research to facilitate their involvement:

‘Finally, consensus meetings will be held with all stakeholders involved including: patients, parents, clinicians and commissioners. The latter will include NHS orthodontic commissioners; these will be apprised of the research at the Delphi stage to facilitate their involvement.’

- List of abbreviations: The is no need of having abbreviations for some sentences that won't be mentioned more that once in the manuscript. It's the case of: BOS; CCT; CENTRAL; CINAHL; CSGBI. So, please remove these abbreviations from the list and from the text.

These have now been removed from the list.

For the abbreviation "MOMENT", it's also written mOMEnt in other places in the text: please write it in one right way throughout the text

Correct amendments of the word are now included in the text.

For RCT, in the paragraph Stage 1, line 12, remove Randomised Controlled Trials (RCTs), let only RCTs, because the abbreviation was already introduced in the text before (the second page of background section, line 3: Specifically, randomised controlled trials (RCTs) in orthodontics published between 2008 and 2012 were analysed....)

Thanks, this has been amended in the text.