Author’s response to reviews

Title: Technology-based functional assessment in early intervention: A pilot study

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REVIEWER COMMENT: Structured summary of method, results and interpretation of results. However, description of pilot study design unclear and implications for future definitive study only partially mentioned. Suggest that some of the main implications are noted in abstract. Perhaps reflect the focus on "technology-based assessment" in the keywords.

RESPONSE: We thank the reviewer for this point of feedback. We have clarified study design (L96) and expanded the conclusions section of our abstract to indicate the types of protocol revisions that were implemented based on results of this study (L111-119). We also have added “technology assessment” as a keyword. We also modified two keywords to more closely align with MeSH terms (L121-122).

REVIEWER COMMENT: p.6, line 169 - Include NINDS in Abbreviations list

RESPONSE: Yes, we agree and apologize for this oversight. We have added NINDS to the abbreviation list (L612).

REVIEWER COMMENT: However, rationale for E-PROs in general could be strengthened. If one considers accessibility issues (e.g. access to PCs) and the fact that self-reported information may not always be comparable to EI providers’ observation of functioning, the researchers must clearly describe the benefits of using e-PROs despite these challenges.
RESPONSE: We appreciate the opportunity to respond to this point of feedback. Family assessment is an established best practice approach to planning and monitoring EI care that provides more information than can be obtained via observation (p. 6, L171-176). E-PROs provide an option for obtaining family input. We agree that the rationale for e-PRO adoption could be strengthened by clearly stating the benefits of a technology-based instrument for access to family engaged EI care (L177-182). We added a reference (ref 12).

REVIEWER COMMENT: p. 8, line 2013 - Include a brief description of EI providers.

RESPONSE: We have included the types of EI providers involved in parentheses (L259-261).

REVIEWER COMMENT: Authors need to provide a clearer explanation of recruitment process, how informed consent was managed, given the feasibility nature of the study. According to Thabane et al. (2010), the informed consent process must include disclosure of purpose of the study, feasibility objectives as well as an explanation of the criteria for success of feasibility.

RESPONSE: We agree and have expanded our description of the consent process in terms of how the study purpose and objectives were communicated to participants (L265-284). We determined success of feasibility to be based on e-PRO enrollment and completion rates being comparable to standard interview-based family assessment methods (50%) and provider perspectives about their ability to screen and enroll families during routine EI service visits (L281-282).

REVIEWER COMMENT: p. 8, lines 221-224 - For predetermined criteria to judge feasibility, the criteria mentioned here seems to focus more on providers' perspective only - i.e. providers' ability to screen and enroll eligible families. Would strengthen findings if participants' perspective is also considered under criteria for feasibility. In Discussion section (p.15) "reasons for study decline" are briefly explored and provides some additional insight.

RESPONSE: We agree and have clarified that our criteria were also based on provider perspective and their ability to screen and enroll families during scheduled EI visits, as well as e-PRO completion rates by families (L298-300). We agree that there is value in also obtaining families’ perspectives but we did not obtain these perspectives in this pilot phase of study. We are obtaining participant perspectives in the current phase of study and have mentioned this as a study limitation (L582-583).

REVIEWER COMMENT: The protocol for the study is described in adequate detail. Additionally, authors should include: -Rationale for participant numbers in pilot study, and focus group discussion schedule should be carefully considered and link to predetermined criteria used to judge feasibility, and -Explain "coaching model" (p.14, line 368). Data analysis for focus group discussion not clearly described.
RESPONSE: Sample size was based on usual care. We know that family assessment enrollment and completion rates are 50% so we estimated that we would be able to enroll 50% or more of the actively enrolled families (L282-283). We have linked the description of provider engagement during and following data collection to the predetermined criteria used to judge feasibility (L290-296). We opted for the use of the ‘primary service provider’ terminology as it is clear in its description of a service approach that places high demand on family involvement during service sessions in the child’s natural environment (L502-503). We provided further detail on our approach to analyzing data from a single focus group (L352-356).

REVIEWER COMMENT: Authors include clear description of how findings will be used to inform the definitive study and the changes to study protocol following the pilot.

RESPONSE: Thank you for this encouraging feedback.

REVIEWER COMMENT: Limitations are clearly stated, sources of potential bias are considered and authors propose strategies to address these in definitive study. It's important to address the cultural diversity issue raised in the discussion to increase enrolment and generalizability of results. Interpretation consistent with objectives and results.

RESPONSE: We appreciate these points of feedback about our work to date.

REVIEWER COMMENT: Please can you make your discussion link better with the background. You make a nice argument for the need for this study in the background, but I can't see how it is picked up and rounded off in the discussion.

RESPONSE: We agree and appreciate the opportunity to improve the coherency of our manuscript. We have attempted to better link our discussion to the background (L536-539, L542-545).

REVIEWER COMMENT: Please remember not all your readers will from the USA. Sometimes you might have to explain what might be country specific terminology a little more than you currently do (e.g., early intervention, early intervention provider, early intervention programme, etc.).

RESPONSE: We appreciate this point of feedback and have done our best to explain early intervention, early intervention provider, etc. (L148-149, L259-261).

REVIEWER COMMENT: It looks like you have cut the number of words down dramatically. One result of this is the loss of many definite articles, i.e. "the". Please could you put them back in, as they make the paper much easier to read.
RESPONSE: We admittedly did make effort to keep the word count within journal guidelines. We appreciate the opportunity to bring back these words that will increase flow. We have done a read-through to re-insert these articles back into the manuscript as best we can.

REVIEWER COMMENT: If you give a little more "background" (i.e. in the background section) about the PROs used in this study, it will make it much easier for the reader to follow the paper.

RESPONSE: We received mixed feedback on the strength of the introduction to communicate the significance of this study. In response to this point of feedback, we attempted to lend more clarity to e-PROs (L177-185, 199-200).

REVIEWER COMMENT: L90 "input" into what?

RESPONSE: We have clarified that e-PROs may provide options for obtain family input on a child’s functioning (L91)

REVIEWER COMMENT: L95 What specifically is an "Early Intervention Provider"? Is it a clinic, is it a person? It isn't entirely clear. What kind of things to they do?

RESPONSE: We opted for the term ‘individual EI provider’ to clarify that we are referring to an individual and not a program or clinic (L96)

REVIEWER COMMENT: L102 "enrolled" into what, the study?

RESPONSE: Yes, exactly. We did not find it necessary to clarify this as it is describing a result, which infers that the reference point is this study.

REVIEWER COMMENT: L105 No need to report to 1 decimal place with a sample size of 37. Just say 60%.

RESPONSE: Sure, we rounded up from 55.9% to 56% per reviewer feedback (L106).

REVIEWER COMMENT: L135 In which country? (it is good to be clear upfront)

RESPONSE: Sure, we have clarified that it was conducted in the United States (L253)

REVIEWER COMMENT: L139 Specify the year that the $1,258 refers to in this text. Just makes it easier on the reader.
RESPONSE: We have specified the year that $1,258 refers to (L 140).

REVIEWER COMMENT: L160 - 170 I think you should briefly mention somewhere in this paragraph that you will describe the PROs in more detail in the methods section.

RESPONSE: We have added that the PEDI-CAT and PEM will be described in further detail in the Methods section (L 170-171).

REVIEWER COMMENT: L171 - 180 I'm afraid I cannot follow your justification for your study you make in this paragraph. It is very dense. Would it be possible to expand and provide some clarifying examples?

RESPONSE: Thank you for this point of feedback. We have attempted to clarify the potential use of e-PROs for patient-centered care planning and outcomes monitoring and research (L 161-174).

REVIEWER COMMENT: L194 - Please could you spell out what kinds of support early intervention programmes provide in the background section?

RESPONSE: We appreciate this point of feedback and have added a sentence to describe the scope of EI service provision (p. 5, L145-146).

REVIEWER COMMENT: L194 - What is an early intervention programme? Is it something a provider implements? It is unclear.

RESPONSE: We appreciate this point of feedback and have added a sentence to describe the scope of EI service provision in the U.S. (p. 5, L145-146)

REVIEWER COMMENT: L195 - Which country? I think many people reading this will know you mean Denver in the USA, but it is good to specify the country.

RESPONSE: Sure, we have included a reference to the country (L196).

REVIEWER COMMENT: L216 "the feasibility of"

RESPONSE: Sure, we have modified the text accordingly (L152)

REVIEWER COMMENT: L217 "conducted by"
RESPONSE: We appreciate the opportunity to further clarify (L289-295).

REVIEWER COMMENT: L226 I don't know what the "10" in superscript refers to. I can't find any footnotes.

RESPONSE: Our apologies for this oversight, we have corrected the formatting of this reference to a citation (L314)

REVIEWER COMMENT: L230 I don't know what the "16" in superscript refers to. I can't find any footnotes.

RESPONSE: Our apologies for this oversight, we have updated and corrected the formatting of this reference to a citation (L318)

REVIEWER COMMENT: L230 -231 The sentence starting "PEDI-CAT domains have…". I would just delete this and say something more general along the lines of how the measure has been found to be both valid and reliable in specific contexts. It isn't really your job to provide the reliability evidence in your paper, just cite the reference for the tool. I think mentioning the ICCs is a little too specific, and makes no mention of the validity of the tool. It doesn't matter if the toll is perfectly reliable if it is not measuring what it is supposed to be measuring.

RESPONSE: We provided references on the validity and reliability of the tool in the introduction (L199-203). We therefore felt comfortable shortening this sentence per reviewer feedback (L318-319).

REVIEWER COMMENT: L242 Why mix medians and means? Non-normal distribution on "home frequency"? If so, maybe say.

RESPONSE: Yes, and we agree about the need to clarify so have done that in the main text (L330)

REVIEWER COMMENT: L246-247 Again, I think you can be a little more general here. Talk about the reliability and validity of the measure in specific contexts as expounded by the reference.

RESPONSE: We have shortened one of the sentences referencing prior studies but have retained the reliability estimates we ran for the study sample (339-340).

REVIEWER COMMENT: L248 This line belongs in the results. Also, do you not need to report this for the PEDI-CAT, if you are going to report it for the YC-PEM?
RESPONSE: We feel that these reliability estimates do not address a study aim per se, but rather are for the purpose of reporting on the psychometric properties of our outcome variables to support Aim 2 analyses. PEDI-CAT raw data are not available to us, just the scores that are automatically generated through the use of the computer adaptive test as administered via iPad. Therefore, we did not report on these estimates for PEDI-CAT. No change made.

REVIEWER COMMENT: L256 Replace "study decline" with "non-participation"?

RESPONSE: We have made this change as requested (L348)

REVIEWER COMMENT: L258 Do you mean "audio-recorded" or did you really use tapes?

RESPONSE: Thank you for this point of feedback. We do indeed mean audio-recorded and have edited the text to make this clear (L351)

REVIEWER COMMENT: L265 Inter "item" correlations between which "items"?

RESPONSE: We modified our wording to clarify the items that were compared (L372)

REVIEWER COMMENT: L267 What "model" you haven't mentioned a model yet. I'm assuming a regression model. Maybe mention you are going to do regression models before this.

RESPONSE: We have clarified this point (L383)

REVIEWER COMMENT: L295 I'm confused as to why n=34 but you collected data on 37 caregivers.

RESPONSE: We only have service use data on 34 of the 37 study subjects. We have made this clear in the footnote for Table 2.

REVIEWER COMMENT: L310-316 This is really interesting!

RESPONSE: Thank you for this encouraging feedback.

REVIEWER COMMENT: L310 I think verbs are missing from the clauses in this line, i.e. "???? a lay summary ...."

RESPONSE: Per reviewer feedback, we have rephrased this for clarity (L350-355)
REVIEWER COMMENT: L318-323 I don't understand what the point of this paragraph is. Please could you make its purpose clearer? Do you not have a table of the full regression model I am guessing you are referencing in line 323? If so please can you present it? Also, what is the relevance of the result that 0-6 year olds were less independent than 12-23 month olds? Isn't that obvious?

RESPONSE: The purpose of the second study aim was to examine if obtaining e-PRO data on functional outcomes (i.e., task performance and home participation) would lend insights about the relationship between EI service use and patient-important outcomes. Due to journal guidelines, we could not include a table depicting the results related to functional task performance. We did clarify that there was no significant effect of EI service use on functional task performance (L449-450).

We were surprised to find that many of the e-PRO scores of functional task performance were within age expected range given that these are children with developmental delays relative to same-aged peers. The purpose of reporting on the trend in 0-6 vs. 12-23 month olds is to show that we detected an age specific trends in some, but not all, PEDI-CAT domains. No change made.

REVIEWER COMMENT: L336 Do you think you need to make these adjustments on a sample size of 37 (or 34, I'm not sure which)? I'm not sure that you do. Although, you could argue this away as "pilot testing" the eventual analyses on a larger sample.

RESPONSE: Yes, this was our approach and we have clarified this (L218-245 and L382-383).

REVIEWER COMMENT: L343 I am unclear on what "unadjusted" means. Are the unadjusted betas regression coefficient while the adjusted betas multiple regression coefficients? If so, please state this. If not, please explain what the "unadjusted" coefficients are. You could perhaps pop in the regression equations for each of the model that were fitted, to make it crystal clear what was fitted.

RESPONSE: The unadjusted models are one-to-one or bi-variate regression models while the adjusted models include multiple covariates. The betas therefore for the unadjusted model reflect the impact of each covariate independently on per child level of involvement, whereas the adjusted model’s betas reflect the change in per child level of involvement while accounting for multiple factors and not simply the one to one relationships. Further, by including the unadjusted results we can then examine if any significant difference in model fit occurs when accounting for an increasing number of covariates in the analysis. We integrated this clarification in the main text (L382-383).

REVIEWER COMMENT: L356 Your uptake was only 44%, please can you comment on how you interpret this as providing evidence that the data collection method is feasible? I think you need to make this clearer.
RESPONSE: We interpret this as the protocol is feasible with modifications and have made that clearer now by providing our criteria for feasibility (with rationale based on usual care enrollment and completion rates of approximately 50%) and provider feedback (L278-284).

REVIEWER COMMENT: L357 Participation in what?

RESPONSE: We have clarified that we are referring to ‘participation in home activities’ (L489-490).

REVIEWER COMMENT: L368 What is a "coaching model"?

RESPONSE: We have changed the wording to match that of the following sentences (“primary service provider model”) for clarity (L501-502).

REVIEWER COMMENT: L382 - 384 I don't understand this sentence. You might be missing a comma, but I don't know as I'm not sure what you are trying to say.

RESPONSE: We agree and have reworded the second sentence for clarity (531-532).

REVIEWER COMMENT: L389 "We were not able to replicate…". You shouldn't be expecting to find this in a pilot study with such low power. This isn't what pilot studies are for.

RESPONSE: We meant that in comparison to prior studies modeling to participation outcomes, we did not find the same effect of environmental support on the outcome. In response to this point of feedback, we have removed “…were not able to replicate recent studies showing…” and replaced it with “…did not find…” (L545).

REVIEWER COMMENT: L407-410 This is not a limitation! This is a pilot study; you were not even trying to do this. You should put these two sentences before the discussion of the quantitative results in order to make this clear to the reader up front.

RESPONSE: We have removed these two sentences from the Limitations section and put them at the beginning of the Discussion section (L484-487).

REVIEWER COMMENT: L424 You say "we have applied these results to inform significant revisions of out sampling and methodology …". I am unclear on exactly what these were, please could you elucidate them more clearly. Perhaps in a table with three columns, i.e. Situation before, evidence for change, changes made, or something along those lines.
RESPONSE: In light of the many edits that have increased manuscript length, and journal guidelines on figure/table limit, we are reluctant to add a table to describe how results have informed revisions to our sampling and methodology. However, in response to this reviewer comment, we have commented on how results suggest that protocol modifications are needed prior to further study and we describe the types of modifications in abstract (L111-119) as well as discussion (L507-527).

REVIEWER COMMENT: Please write your aims and expand the specific objectives to be addressed clearly at the end of the introduction.

RESPONSE: We have modified the aims to be more clear (L215-246).

REVIEWER COMMENT: Please give a little more detail about the setting (eg is Denver Metro a hospital?) with dates of study.

RESPONSE: We have specified that Denver Metro is an area in Colorado in the United States (L252).

REVIEWER COMMENT: How were the patients recruited and consented? And by whom?

RESPONSE: We have clarified that EI providers recruited participants using flyers and that participants consented and enrolled in the study using their personal computers or iPads provided to participants by EI providers (L264-267).

REVIEWER COMMENT: Please add how focus group data were qualitatively analysed in methods section.

RESPONSE: We provided further detail on our approach to analyzing data from a single focus group (L351-355).

REVIEWER COMMENT: Please provide confidence intervals rather than SEs in the text of the regression results on page 14.

RESPONSE: We have updated Table 3 to include the Confidence Intervals, however we also want to note that that SEs may be more appropriate than confidence intervals given the small sample size. Confidence intervals, particularly in small samples, tend to be imprecise measures that gave us pause in including them over standard errors.

REVIEWER COMMENT: Please write abbreviated terms M, Mdn in full in the text.
RESPONSE: Yes, we have made this edit throughout the main text (L440-460)

REVIEWER COMMENT: Please add a cautionary caveat that the regression (hypothesis testing) results should be interpreted with caution because of small sample size.

RESPONSE: We agree and have included this statement in discussion (L542) and limitations (L574)

REVIEWER COMMENT: Please add a rationale for the chosen sample size before data analysis section. A calculation does not need to be performed but some rationale would be helpful.

RESPONSE: We agree and describe how sample size was determined based on an educated guess, given what we know about enrollment and completion of family assessment in EI through usual care using interview methods as described in the background. We provide this rationale in methods (L280-283).

REVIEWER COMMENT: In Table 1 please list amount of missing data in the footnote eg Missing data: Family income (1) - there does not appear to be missing data for child race/ethnicity variable even though asterisked.

RESPONSE: We agree and have made these edits in Table 1.