Author’s response to reviews

Title: A cost-effectiveness study of ICT training among the visually impaired in the Netherlands

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Author's response to reviews:

BMC Ophthalmology

Editorial Office

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Subject: Submission of revised paper ‘A cost-effectiveness study of ICT training among the visually impaired in the Netherlands’ BOPH-D-17-00245

Dear Dr. Tu,

Thank you for your email [25th of September 2017] enclosing reviewers’ comments. We have carefully reviewed the comments and revised the manuscript accordingly. On the following pages, you will find our responses in a point-by-point manner, with clear indications of amendments made to the original manuscript.

On behalf of my co-authors, we thank you for the opportunity to resubmit the paper and we hope that the revised version of the manuscript is suitable for publishing. We look forward to hearing from you again.
Yours sincerely,

Nathalie Patty, MSc

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Response to Reviewer 1:

Thank you for our critical remarks on our manuscript. We appreciate your time and effort for reviewing our manuscript.

Reviewer 1, point 1: Abstract line 7 (and elsewhere eg page 6 line 13): It is confusing to refer to the several different instruments used as outcome measures as if they were a single questionnaire: it should be made clear that at each relevant time point there were 6 questionnaires employed. Was the order of the 6 questionnaires the same on each occasion?

Response: We agree with the reviewer that this could be more clearly described in the manuscript. The questionnaire used at each relevant time point consisted of six separate questionnaires; the order of the six questionnaires was the same on each occasion. We have adjusted the manuscript. Amendments can be found in the section ‘Abstract’, Line 9 to 10, Page 2 and in ‘Methods’, Line 3 to 4, Page 7. In the section ‘Methods’, Line 4, Page 7, we also point out that the order of the six questionnaires was the same each measurement time.

Reviewer 1, point 2: Page 4 lines 2/3: In an international journal, it would be better to give VA in logMAR notation.
Response: We agree with the reviewer that it is better to give VA in logMAR notation. The amendments can be found in the section ‘Background’, Line 3 to 4, Page 4.

Reviewer 1, point 3: Page 4 line 19: There is another study of cost-effectiveness of low vision rehabilitation published recently - Bray et al Acta Ophthalmologica doi:10.1111/aos.13255

Response: Thank you for pointing out this article, it is nice to see that studies that are more recent have been conducted about the cost-effectiveness of low vision rehabilitation. We have added this in our article. Amendments can be found in section ‘Background’, Line 20, Page 4 and Line 8 to 10, Page 5, in the ‘Discussion’, Line 14 to 18, Page 16 and Line 9 to 10, Page 17.

Reviewer 1, point 4: Page 6 line 3: I appreciate the rationale for the "before and after" study design, even though it is not desirable. However I think the recruitment of participants should be reported with as much rigour as if it was an RCT. How many participants formed the potential pool? How many were interested? In how many cases was it judged inappropriate? How many had training but were not in the study? How many dropped out of the training? This information could be added to the Appendix 1 figure, which is currently rather uninformative, and this should be moved to the main text. The authors need to disclose as well as they can any sources of bias in the recruitment/follow-up, or ways in which they attempted to avoid bias. I was dismayed to see that researchers in some cases helped to complete the questionnaires with participants, which is not good scientific practice. It appears there was no age limit on the participants (based on Table 1) - so why use ICECAP O rather than ICECAP A? Was there a target number of participants based on a power analysis? Or recruitment across a fixed period? Were participants undergoing any other forms of treatment or rehabilitation at the same time?

Response: For the sake of clarity, this chunk of questions has been broken down into four parts.
Part 1. Reviewer states “Page 6 line 3. I appreciate the rationale for the "before and after" study design, even though it is not desirable. However I think the recruitment of participants should be reported with as much rigour as if it was an RCT. How many participants formed the potential pool? How many were interested? In how many cases was it judged inappropriate?”

Response: We believe that this needs further explanation. Annually 180 people received ICT training, not simultaneously with other training. All interested people who met the inclusion criteria were included in the study. Amendment can be found in section ‘Methods’, Line 19 to 21, Page 5.

Part 2. Reviewer states: “How many had training but were not in the study? How many dropped out of the training? This information could be added to the Appendix 1 figure, which is currently rather uninformative, and this should be moved to the main text. The authors need to disclose as well as they can any sources of bias in the recruitment/follow-up, or ways in which they attempted to avoid bias. I was dismayed to see that researchers in some cases helped to complete the questionnaires with participants, which is not good scientific practice.”

Response: We fully agree with the reviewer that more information about dropouts should be mentioned in the manuscript, reasons for participants dropping out, etc. We have therefore adjusted the Appendix figure 1 by adding the dropout rates and reasons for dropping out in the figure. We have also decided to move this figure to the main body of the manuscript as we agree with the reviewer that this information is important and should therefore be more visible. This figure has been named “Figure 1. Flowchart of enrollment process, participant dropouts and timeline for the measurements” and can be found in the section ‘Methods’ on Page 6. Amendments have also been made to the manuscript. These can be found in section ‘Methods’, Line 18 to 19, Page 6 and in the section ‘Results’, Line 15 to 16, Page 9.
We also acknowledge that it is important to disclose any sources of bias in the recruitment process and the follow-up, as well as attempts made to minimize bias. We would like to point out that in clients were eligible and invited to participate if they were not simultaneously receiving any other training, as mentioned in section ‘Methods’, Line 20 to 22, Page 5. We do recognize that trainers who recruited the clients could have been a potential source of bias. However, trainers were instructed to invite every new potential participant. Amendment has been made in section ‘Method’, Line 21, Page 5.

Furthermore, we agree that it is not good scientific practice to help respondents filling in questionnaires. However, we would like to point out that the majority of the participants were elderly (>63 years old) and many of them lived alone and struggled with basic daily activities (at baseline 38% of the participants received some form of home healthcare). In some cases, when enrollees struggled filling in the questionnaire, and did not have anyone who could help them we assisted through telephone, by literally reading the question and writing down the answer that the respondent would provide. This has been justified in the manuscript under section ‘Methods’, Line 16 to 17, Page 6.

Part 3. Reviewer states: “It appears there was no age limit on the participants (based on Table 1) - so why use ICECAP O rather than ICECAP A?”

Response: This is a highly relevant question. The decision to use ICECAP-O was made because we were informed by the ICT trainers that the majority of the people receiving ICT training were elderly. The analysis of the data at baseline supported the choice of using ICECAP-O instead of ICECAP-A, as the mean age at baseline was 63 years. Our explicit choice of using ICECAP-O has been specified in the manuscript. Amendment can be found is section ‘Methods’, Line 17 and 20, Page 7.
Part 4. Was there a target number of participants based on a power analysis? Or recruitment across a fixed period? Were participants undergoing any other forms of treatment or rehabilitation at the same time?

Response: These questions are highly important. The number of participants was not based on a power analysis, as the size of the treatment effect and its variance was not known. Therefore, a power calculation would not have been possible. To limit the burden for the organizations, we decided to limit the inclusion period for seven months, this is mentioned in the manuscript section ‘Methods’, Line 19, Page 5. Those who participated were only eligible to be a part of the study if they were not simultaneously receiving any other rehabilitative training that could bias the outcome. This is mentioned in the manuscript section ‘Methods’, Line 20 to 22, Page 5. Participants underwent normal eye care (see the result of medical consumption) and they did not receive other training than ICT training at the same time.

Reviewer 1, point 5: Page 6 line 13: The choice of questionnaires is not well justified. This is particularly the case for the questionnaires which are not available to the reader to go and look up (page 7) - although citations are given, the text is either not available or in Dutch (refs 8,9). It appears from information given in the Results, that "medical consumption" also includes what might more usually be termed "social care" ie, assistance around the home. What was the hypothesis about change in "Medical Consumption" or "Productivity Cost"? What use was made of the information gathered on level of education or daily activities (I don't really know how to interpret "work disability" and "pre pension"?)?

Response: For the sake of clarity, these questions have been broken down into three parts.

Part 1. Reviewer 1 states: “Page 6 line 13. The choice of questionnaires is not well justified. This is particularly the case for the questionnaires which are not available to the reader to go and
Response: We agree with the reviewer that the choice of the specific parts of questionnaire could be better justified. We have highlighted that the potential improvement of computer skills, as measured by the DAI is the vehicle towards a possible improvement in health related quality of life (EQ-5D) and well-being (ICECAP-O). We have adjusted the manuscript accordingly. Adjustments can be found on in section ‘Methods’, Line 9 and 10, Page 8. As for the references, we apologize for reference # 8 (the reference for Medical Consumption, currently reference # 9). This questionnaire and the instruction manual (which is the source of reference # 9) is unfortunately only available in Dutch and upon request. Access to view the questionnaire can be done directly contacting imta through their website (https://www.imta.nl/contact/) or by contacting kanters@eshpm.eur.nl. Source # 9 has been updated please see ‘References’, Page 22, Source number 9. Furthermore, we want to point out that the original source # 10 (referred to as source #9 by the reviewer) describing the Productivity Cost questionnaire is available in English and online. This source is can be found under source 10 in the list of References on Page 22. Access to view the questionnaire can be requested by contacting imta directly through their website (https://www.imta.nl/contact/) or by contacting kanters@eshpm.eur.nl.

Part 2. “It appears from information given in the Results that "medical consumption" also includes what might more usually be termed "social care" ie, assistance around the home. What was the hypothesis about change in "Medical Consumption" or "Productivity Cost"?

Response: We believe that the rationale behind including “Medical Consumption” and “Productivity Costs” needs some further clarification. We would like to point out that it could not be ruled out that ICT training may have an effect on medical costs and productivity costs. For example, there may be a risk (although we think minimal) that ICT training could increase or decrease medical consumption. Likewise, ICT training could also result in lower productivity
costs. Only solid proof can justify leaving out these costs. Therefore, according to standard economic evaluation, from a societal perspective, these costs should not be disregarded without solid proof that they have no effect. We also like to point out that both of these questionnaires have been validated. Furthermore, we did not have a specific hypothesis regarding the change in Medical Consumption or Productivity Cost.

Part 3. What use was made of the information gathered on level of education or daily activities (I don't really know how to interpret "work disability" and "pre pension")?

Response: We believe that this part needs some further explanation. As for the information gathered on the level of education or daily activities, it is important to know the composition of the sample. It can be assumed that the level of education will have an effect on the total training time required as well as how severe the visual impairment is. For example, a high-educated person may learn faster and, hence, may not require an extensive ICT training or may not be as severely impaired, and therefore capable of completing a higher education level opposed to a person who is severely impaired. Similarly, daily activities can also be assumed to be effected by the severity of the visual impairment. Concerning the terms "work disability" and "pre pension”, we have adjusted the terminology. Amendments can be found in Table 1. The term ‘(pre) pension’ has been replaced with the word ‘early retirement’ and the term ‘work disability‘ has been replaced with ‘Incapable of working due to sickness or disability’.

Reviewer 1, point 6: Page 8 line 1 (and p11 line 9): Is the data parametric or non-parametric? This should be established and then either t-test or Wilcoxon signed rank performed as appropriate.

Response: We believe that this needs some clarification. ICECAP-O (including VAS), EQ-5D (including VAS) and CarerQol (including VAS) are all parametric measures due to their level of
measurement and as the sample size is above 30 (n=45 and n=38), hence a normal distribution can be assumed. Please see amendment in section ‘Methods’, Line 22, Page 8. However, we also conducted a non-parametric test to validate the results. This is mentioned in the manuscript; see section ‘Methods’, Line 22 to 23, Page 8.

Reviewer 1, point 7: Page 8 line 8: What is the rationale for including all the medical costs? Page 12 suggests it was because they proved to be higher after training, but the authors couldn't have known this at the start.

Response: Thank you for your critical remark. As previously mentioned (see comment Reviewer 1, point 5, response ‘Part two’), we would like to point out that it cannot be ruled out that ICT training may have an effect on medical costs. For example, there may be a risk (although we think minimal) that ICT training could affect medical consumption. Therefore, according to good HTA-practice, from a societal perspective, these costs should be taken into account.

Reviewer 1, point 8: Page 8 line 14: I think more detail is needed on drop-outs, or those who had not completed training. These people still appeared to create some costs, and yet these are not accounted for. You say some people had not completed training "within the study period". Did this mean that their training was particularly lengthy, and therefore more costly? If they are not being counted this would artificially lower the mean training cost.

Response: We agree with the reviewer that more detail should be provided about the dropouts (those who did not complete the training). Therefore, we have adjusted Appendix 1 by including the dropout rates and by moving this figure in to the main body of the manuscript. Figure 1 can be found in the section ‘Methods’, Line 20, Page 6. Amendment can also be found in the section ‘Results’, Line 15 to 16, Page 9. However, we believe the question the reviewer presents regarding the costs of the training needs further clarification. In the analysis, the costs generated
by participants who had not completed the training were not taken into account. This may produce a small underestimate in the costs. On the other hand, likewise, participants who did not complete the training may also have experienced positive effects on their well-being, which was also not taken into account.

Reviewer 1, point 9: P11 line 9 (and Table 2): p<0 and p=0 are impossible - presumably you mean p<0.001?

Response: We apologize for the typo. We have adjusted the p-value correctly. Amendment can be found under ‘Impact of the ICT training’, Line 9, Page 12 and in Table 2, Page 13.

Reviewer 1, point 10: P13: Are the QALY estimates based on the change in the EQ-5D scores? How is the "years of well-being" derived from the ICECAP score? I thought that the metric in the ICECAP was "Years of Full Capability"? Although the ICT training effect might persist for 15 years (although that's a bit of a stretch considering there is only 3 months of follow-up data to base it on), it seems very unlikely that the health status of the participant will be maintained as they age another 15 years.

Response: We apologize for not being clear on what we base the QALY estimates. The QALY estimates are based on the change of EQ-5D scores. We have adjusted the manuscript so that it is clear on what the QALY estimates are based on, as well as the ‘years of well-being’. Amendments can be found in the section ‘Methods’, Line 5 to 7, Page 9.

Furthermore, the metric for ICECAP-O is indeed ‘Years of Full Capability’. However, as the capability approach defines the well-being in terms of what an individual is capable to ‘be’ or ‘do’ (for further information regarding ICECAP–O see source # 8 in the manuscript), the
decision was made to use the term ‘years of well-being’ instead of ‘years of full capability’. We have highlighted our choice regarding the terminology in the manuscript. Amendments can be found in the ‘Methods’, Line 11 to 17, Page 7.

Our baseline assumption is persistent effects for 10 years. See section ‘Cost–effectiveness’, Line 3 to 4, Page 14. The 15 years is an upper limit in our sensitivity analysis and might be an optimistic one, as suggested by the reviewer. We do want to point out that we do not assume that the health status will remain the same in 10 years, what is being assumed is that the gain in health status will last for 10 years.

Reviewer 1, point 11: P14 lines 12/13: I don't understand this sentence?

Response: Amendment has been made in the section ‘Uncertainty analysis’, Line 12, Page 15. When only the costs of ICT training were included (excluding the change in medical costs), the results of the bootstrapping analysis indicated that in 99% of the cases, well-being was gained with limited additional costs.

Reviewer 1, point 12: P16: The discussion about the need for a more sensitive measure than EQ-5D is highly relevant, although I would expect the authors to consider the use of VisQoL, considering that it was designed for this purpose.

Response: We agree that the outcome VisQoL is highly relevant within visual rehabilitation, as it is a utility measure specifically designed for health economic evaluation of eye care and rehabilitation programs. However, our focus was in the general and generic dimensions of health related of quality of life and capabilities that could be influenced by better ICT skills. Those
dimensions can be found on ICECAP-O and in the EQ-5D. However, we agree with the reviewer that the VisQOL might also pick up improvement in these skills.

Reviewer 1, point 13: P16: Although this is primarily a cost-effectiveness study of what is described as an established intervention, I think some discussion of the intervention itself is warranted. Firstly I thought the direct outcomes of the training (improvement in computer use) were rather disappointing as shown in Table 1, with substantial proportions of participants still having difficulty with many basic aspects of operation (e.g., screen and mouse). The training protocols are obviously also very loosely interpreted, since individual programmes ranged from 2-63 sessions. One could argue that this training protocol needs to be more tightly specified before meaningful economic evaluations can be carried out.

I also thought the authors might have made more of the result of the caregivers questionnaire, which showed the "delayed" effect from T1 to T2, rather than immediately from T0 to T1. This would seem to be much more promising as a sign that participants were further developing their usage, and independence, in the period after training.

Response: We appreciate the critical marks made by the reviewer. We agree that a more tight protocol would have improved the internal validity. However, at the expense of the external validity, because in reality the number and content of the sessions varies as the training is tailored to the individual client’s needs. This study intends to analysis the cost-effectiveness of the current practice in the Netherlands, including all its particular individual variations.

Reviewer 1, point 14: Typos Table 2: 77.02 in penultimate row

Response: Thank you for your accuracy. We have corrected the typo. Amendment can be found in Table 2, Page 13.
Response to Reviewer 2:

Thank you for our critical remarks on our manuscript. We appreciate your time and effort for reviewing our manuscript.

Reviewer 2, point 1: The authors have not used a control group. They justify this decision by saying that this would have been unethical and impossible. I agree that this would be true if every individual receives ICT training. However, on Page 6 (line 3) the authors suggest that only individuals who were interested in ICT training were enrolled in the study. This would suggest that there were a group of individuals who were not interested in ICT training and could therefore form part of a control group. Why was this not considered?

Response: The reviewer is correct in pointing out that patients who were not interested in ICT training could have formed a control group. However, patients who are not interested in ICT training probably have sufficient ICT competence, so not in need of training, or are not motivated in enhancing their skills. Both groups would probably be quite different and, hence, be incomparable to our target group. As a result, we did not see good opportunities for a valid control group, apart from our ethical arguments.

Reviewer 2, point 2: Some information needs to be provided about subjects who did not complete the training during the study period and why this was the case as this has implications for costing. It is also not clear how many participants actually enrolled on the study and what the drop-out rate was and reasons for drop out (I note the authors mention health related or personal reasons). This also needs to be clarified as this provided important information which is relevant to the study as well.

Response: We acknowledge that the dropout rate should be reported as well as the reasons behind the dropouts. To clarify this we have adjusted Appendix 1 by adding the dropout rates in
the figure. We have also moved Appendix 1 to the main body of the manuscript. Appendix 1 is now ‘Figure 1’ and can be found in the section ‘Methods’ on Page 6. Furthermore, minor adjustments can also be found in the manuscript section ‘Results’, Line 15 to 16, Page 9.

Reviewer 2, point 3: The authors use questionnaires some of which are in Dutch and some of which are in English? It is not clear whether a mixture of languages were used or the English questionnaires were translated into Dutch. This should be made clear.

Response: We agree with the reviewer that this needs to be clarified. The questionnaires (which was composed of six questionnaires) were all in Dutch, and as the study took place in the Netherlands, the respondents received the questionnaire in Dutch. To clarify this important piece of information, we specify this in the manuscript under section ‘Methods’, Line 20 to 21, Page 7.

Reviewer 2, point 4: Abstract: Line 4: 'costs and effects' - what do you mean by effects - this needs to be clarified Line 7: More detail about participants is needed. How many participants took part in the study? What was their mean age?

Response: We admit that ‘costs and effects’, as stated in the abstract may be confusing for the reader. By ‘costs and effects’ we mean ‘cost-effectiveness’. We also agree that some basic information should be mentioned in the Abstract such as how many participants participated and their mean age. Amendments can be found in ‘Abstract’, Line 4 and 8 to 10, Page 2.

Reviewer 2, point 5: Background: Page 4 Line 3 & 4: Please use LogMAR notation.

Response: We agree with the reviewer that it is better to give VA in logMAR notation. The amendments can be found in the section ‘Background’, Line 3 to 4, Page 4.
Reviewer 2, point 6: Methods: Page 5: Line 21: What do you mean by the waiting time for enrolling in the training was limited?

Response: We agree with the reviewer that the sentence is not clear. In this sentence we mean that the waiting time for being enrolled in the training was short. The sentence has been adjusted and the amendment can be found in the section ‘Method’, Line 2, Page 6.

Reviewer 2, point 7: Methods: Page 6 Line 7-12. Be explicit about what the first, second and last questionnaires are? It sounds like you only used one questionnaire where as in reality you used several questionnaires at each point in time. This section probably needs to be reworded to make it clear.

Response: We agree with the reviewer that this section needs to be more specific. We have adjusted the manuscript. We used one questionnaire, which was composed of six questionnaires. Amendments can be found in the section ‘Methods’, Line 3 to 4, Page 7.

Reviewer 2, point 8: Results: Some information about the eye disease causing visual impairment would be useful. Include this information in table 1.

Response: We believe that this point needs some further explanation. The aim of our study is to evaluate the cost-effectiveness of ICT training of the current practice in the Netherlands. We focus on the participants’ disabilities with the aim to evaluate how these disabilities could improve through ICT training, in terms of health related quality of life as well as well-being. Thus, our focus is not based on the medical reason causing the participants’ disability. Therefore, adding information about eye disease would not add value to this study.
Reviewer 2, point 9: Results: Page 11 Line 3 and 4: The domain 'security' also improved in line with 'enjoyment' and 'control'. Why was this not mentioned? It is mentioned on the previous page.

Response: We thank the reviewer for being accurate. On page 10 we only describe the direct changes that was seen between T0 and T1. On page 11, we describe outcomes of the paired t-test. We have added the outcome of the t-test for the domain ‘security’ as we indeed mention on page 10 that this was one of the most important domains when comparing the outcomes. Furthermore, when conducting the t-test for the domain ‘security’, although non-significant, it was one of the most substantial improvements. Amendments can be found, in section ‘Impact of the ICT training’, Line 3 and Page 12.

Reviewer 2, point 10: Results: Page 12: Does the effect of ICT training persist? - Why was a one way ANOVA not carried out? Why use two separate t-tests? Did you consider Bonferroni’s adjustment as you have several t-tests?

Response: We appreciate the reviewer’s critical point of view. However, we would like to point out that a one way ANOVA is appropriate to use when one wants to determine whether there are any statistically significant differences between the means of three or more independent (unrelated) groups. As our outcomes are based on one group of participants with three dependent measurement moments, an ANOVA would seem not to be the most appropriate test. As for the Bonnefori’s adjustment, we did not consider Bonferroni’s adjustment as the number of t-tests compared to number of observations is still limited and the reader can use a stricterF p-value (lower than 0.05) if necessary. Moreover, conform the recent advice of the American Statistical Association (http://www.amstat.org/asa/files/pdfs/P-ValueStatement.pdf), researchers should focus less on p-values, and more on the relevance of coefficients (e.g. the magnitude of the gain in well-being and/or quality of life).
Reviewer 2, point 11: Discussion: Some mention should be made about why difficulties with mobility increased after training? Also why does 'pain' and 'discomfort' decrease after ICT training?

Response: We agree with the reviewer that we should elaborate more on this issue. However, we can only speculate on the reasons of these changes. It could perhaps be possible that better ICT skills promoted confidence for participants to move more freely in their environment. However, when doing so, the participant may have also realized that they actually are less mobile than what they had initially thought. Similarly, the score of ‘pain / discomfort’ decreased, probably as daily activities became easier to perform after having acquired better ICT skills.