Author’s response to reviews

Title: Development of an inventory list of goals using the International Classification of Functioning, Disability and Health in a population of non-ambulatory children and adolescents with cerebral palsy treated with botulinum toxin A

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Author’s response to reviews:

Dear BMC Pediatrics Editorial Office,

I want to thank the reviewers and editor for their valuable comments on our manuscript. I am writing to resubmit for publication the revised version of the manuscript titled: “Development of an inventory list of goals using the International Classification of Functioning, Disability and Health in a population of non-ambulatory children and adolescents with cerebral palsy treated with botulinum toxin A” (BPED-D-16-00453). Please find below a point-by-point response to the comments.

Editor Comments:

1. STROBE Checklist

A completed STROBE checklist for observational studies is attached as an additional file, and each item was referenced with a page number in the manuscript. Some additional statements were provided in the manuscript to follow the STROBE guidelines.

2. Additional files

A section “Additional files” (after the References/Figure legends) is provided. There are two additional files included in the manuscript. The file name, title of data, and description of data have been provided for each additional file.
3. Availability of data and materials

All data generated or analyzed during this study are included in the manuscript. The raw data can be requested from the corresponding author.

Reviewer #1 Comments

Major Comments:

1. Description of participant characteristics and history

a) How many parents/opinions were included for each child?

The interviews allowed for both parents to be interviewed for each child. However, there was only one parent interviewed for each child who indicated that he or she made the healthcare decisions as the primary caregiver.

The following sentence was included under “Study Design”: “Both parents were invited to participate in the interview.”

The following sentence was included under “Results, Involvement of parents in goal setting”: “For each child, there was only one parent identifying the goals who was considered to be the primary caregiver by the family.”

b) Was BoNT-A being recommended for immediate use at the time of interview?

A few sentences to describe this process has been included under the section of “Study Design”: “The interviews took place after the assessment by the therapist, but before a decision was made with the physician. After a decision was made and BoNT-A treatment was recommended, it was administrated either in clinic or under sedation at a different time.”

c) Had they previously consented to BoNT-A injections for their child, and how many times?

d) What was the outcome of previous BoNT-A injections?
We agree with the reviewer, and have created an extra table to describe the number of BoNT-A injections the child received before and after the interview, the changes after BoNT-A injections, and the overall outcome.

We have included Table 3 and a paragraph to describe the frequency and outcomes of BoNT-A injections in “Types of goals raised by parents” in the Results section:

“The changes that were observed in each child after the interview and administration of BoNT-A treatment are described in Table 3. Most children received BoNT-A treatment in the lower extremity. There were 15 parents (75%) who described that there were positive outcomes for their child after BoNT-A treatment. Parents described outcomes related to body structures and function, such as increased range of motion and reduced tone, as well as, outcomes related to activity, such as ease of dressing and personal hygiene care. Two parents identified participation outcomes, including horseback riding and swimming. At follow-up visits, these outcomes were helpful in identifying whether parents’ identified goals were achieved and for future goal setting with healthcare professionals.”

2. Addition of goals to the initial inventory produced by the national botulinum toxin conference seems to have been contributed by healthcare professionals involved in the study. Were they co-authors on the paper? If they were not, they should be considered as participants in the study, and more details on them should be included. Some relevant details may include the number of years they have been in practice, the type of practice they have, whether they were commenting on the specific subjects/children with CP in the study or commenting in general based on their experience, whether they know the study subjects, and how many were included.

The reviewer is correct that the healthcare professionals on the Spasticity Management team have contributed to the inventory of goals. They are not co-authors, but they are acknowledged in the manuscript. There was one nurse, two physiotherapists, one occupational therapist, and one child life specialist. Each healthcare professional has more than five years of experience on the Spasticity Management team. These healthcare professionals were commenting on the general goals in the inventory.

We have added a description about the team of healthcare professionals in the “Development of the inventory of goals” in the Methods section:

“The initial inventory of goals was provided to the healthcare professionals at the Spasticity Management Clinic for feedback. The team of healthcare professionals included one nurse, two physiotherapists, one occupational therapist, and one child life specialist with over five years of experience. The healthcare professionals were also asked for the additions of new goals that were
missing from the inventory, phrasing and clarity of the goals, and the categorization of goals in the ICF framework.”

3. A link is not made between the inventory of treatment goals and the outcomes of BoNT-A treatment. It would be helpful to describe this either in the Background or in the "Development of the inventory goals" section of the Methods. In other words, is there evidence that the goals are realistic and match potential outcomes after treatment or are they generic goals that parents have for their child that are not specific to BoNT-A treatment?

We included the following information to clarify the link between the inventory of treatment goals and the outcomes of BoNT-A treatment in the “Development of the inventory of goals” section of Methods:

“The goals were specific to children and adolescents with CP and receiving BoNT-A treatment. Some of these goals were identified by parents in a retrospective study and achieved in 75-80% of cases [16].”

The reviewer raised an interesting point, but the focus of this study was to explore parents’ goals. We would agree that we would link the goals with potential outcomes after BoNT-A treatment, but this could be conducted in future studies.

4. Table 3 is missing

Our apologies for not attaching Table 3, which was missing in the original submission of the manuscript. There are now three tables included in this manuscript. Table 1 describes participant characteristics. Table 2 (originally stated as “Table 3”) provides all of the treatment goals that were identified by parents following a discussion with healthcare providers. Table 2 is included as an attachment in the revised manuscript submission. The new Table 3 is the description of each BoNT-A injections in each child.

5. To show that the inventory can be helpful during the goal setting process (which is the conclusion of the manuscript), it would be important to systematically document the initial parent responses to the open-ended question used in the study design: "What kinds of goals do you have for your child with cerebral palsy after receiving botulinum toxin treatment?" Otherwise, as the study is reported currently, it is difficult to quantify how much the inventory helped in the process. [Also, it may be useful in the future to determine the parents' perceived usefulness of the inventory, and the effects of inclusion of the inventory on a
The reviewer raised a good point about the usefulness of the inventory of goals, both by the number of goals raised by parents and the perceived partnership between families and healthcare professionals. The focus of this study was on the development of the inventory of goals rather than the perceived usefulness of the inventory.

Based on the study design, we did not audio-record the interview and cannot provide a systematic document of parents’ responses to the initial open-ended question. However, the types of goals raised by parents were mainly body structure and function goals, such as reduced muscle tone and improved range of motion, as described in the Results section. The inventory of goals was helpful in prompting parents to consider activity and participation goals. As stated in the Results section, “most parents did not mention goals related to participation until they were presented with the inventory list of goals.” Although it is difficult to quantify the extent to which the inventory of goals helped in the goal-setting process for parents, the inventory of goals can be used as a resource for parents to consider different types of goals, including body functions and structure, activity, and participation.

We agree with the reviewer that it would be helpful to determine parents’ perceived usefulness of the inventory and included this information in the “Implications for future research” under the Discussion section.

“A future study is needed to conduct qualitative interviews in order to better understand parents’ past experiences of BoNT-A treatment, the information parents may require about setting relevant and meaningful goals during treatment, and parents’ perceived usefulness of the inventory of goals.”

Minor Comments:

1. We also agree that “range of motion of muscle limbs” can be reworded to “joint range of motion”, and have made the change in the manuscript (Page 3, line 12).

2. Table 1 – identify whether, where, and how frequently BoNT-A was used in the past.

We agree with the reviewer, and have included the frequency of BoNT-A injections before and after the interview in Table 3 in the “Types of goals raised by parents” under the Results section.
3. To ensure consistency, we also agree with the reviewer that parents will be referred to as “participants” and children and adolescents will be referred to as “subjects”. We have made the changes in the section of “Subject Characteristics.”

4. A description of how cognitive impairments was assessed is provided under the section of “Subject Characteristics”:

“Cognitive impairments were assessed informally by the healthcare professional team. Children and adolescents were identified to have cognitive impairments based on their level of understanding during discussions in clinical appointments, such as their processing level of information, responsiveness to questions, and communication with healthcare professionals.”

5. We agree with the reviewer that the goals were “among parents” of children with CP, and have made the change on page 5, line 40-41.

6. We agree with the reviewer and inserted the word “and” after the word “child” on page 5, line 58.

Reviewer #2 Comments

1. Methodology aspect

We agree with the reviewer that this study followed a cross-sectional observational study design, in which information was collected from participants at a single time point. We have made the changes from the word “prospective” to “cross-sectional” in the sections “Abstract Methods” and “Implications for clinical practice”.

2. There is no need for control group in this study since it is not applicable with the research.

We also agree that there is no need for a control group in this study since it is not applicable with the research.

3. Consideration as a research article
According to BMC Pediatrics, research articles report on “original primary research” and the main findings of the study results. As such, we report on the findings from a convenience sample of 20 parents and the types of goals that they have for their child after BoNT-A treatment.

4. Application of the inventory of goals for botulinum toxin injections among children and adolescents with CP

Clinicians/healthcare professionals need to guide parents whether added or chosen inventory goals are realistic to patients functional capacity or not, which in this study involved GMFCS IV and V.

This comment is similar to the third comment from Reviewer 1 in whether there is evidence that the goals are realistic and achievable after treatment. We agree with the reviewer that presenting an inventory of possible goals, such as using a walker and participating in sports, could raise parents’ expectations in considering BoNT-A treatment. We hope that healthcare professionals can use the inventory of goals with parents as a springboard to help guide and inform parents about setting appropriate goals and individual counseling.

We included a sentence under “Implications for clinical practice” in the Discussion section: “Although presenting an inventory of possible goals could potentially raise expectations in parents considering BoNT-A treatment that may need to be managed by healthcare professionals.”

Additional Note

There is a discrepancy for Figure 2. The figure indicates that there was one new goal that was added from parents. However, there were two new goals added.

The manuscript text reads: “The parents identified sleep with few disturbances as a body structure/function goal and sit comfortably with good posture as an activity goal that were missing from the inventory.” Therefore, there is a discrepancy between the figure and the text. We have revised the figure to indicate that there were two new goals added from parents.

Some grammatical changes were corrected throughout the manuscript.

On behalf of my co-authors Linda Nguyen and Dr. Ronit Mesterman,

Kind regards,

Jan Willem Gorter MD PhD FRCP(C)

Professor Pediatrics, McMaster University